

**Evaluation of health and social
care provision for ventilator-
dependent children in the UK:
Costs and outcomes**

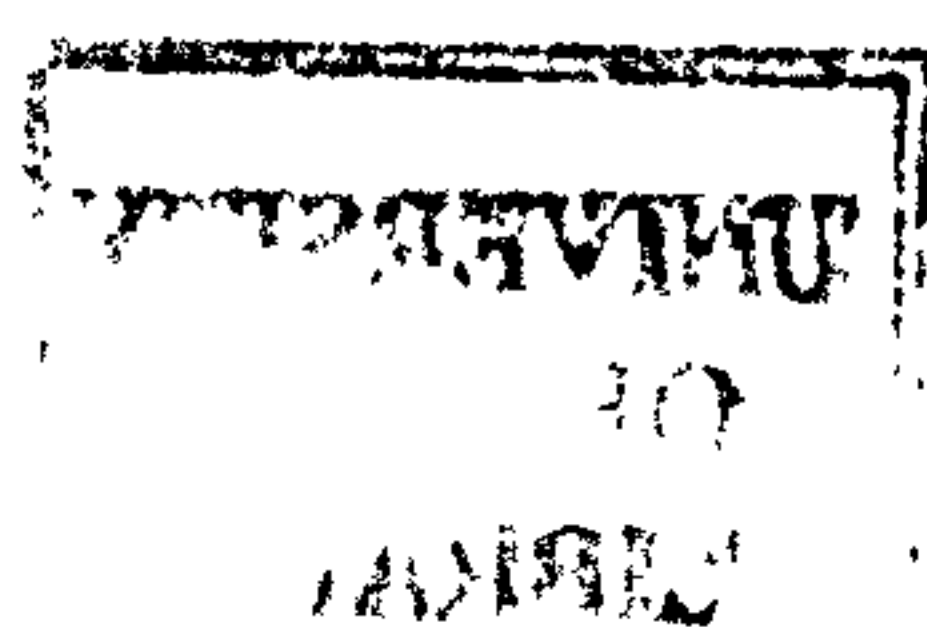
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Submitted for the Degree of Doctor of Philosophy
in Health Sciences

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2004



Abstract

The emergence of a new population of ventilator-dependent children has presented both the National Health Service and social care providers with considerable challenges in terms of commissioning cost-effective and child-centred services that have a positive impact on the health-related quality of life of children and their families. Doctors and commissioners of services have been hampered in their decision-making by the lack of an evidence base concerning this low volume high cost population of children.

This thesis contributes to the evidence base concerning ventilator-dependent children by generating and synthesising new quantitative and qualitative knowledge around 4 key research themes, including:

1. The origins and size of the population
2. The health-related quality of life of children
3. The costs of care, and
4. Satisfaction with services.

In terms of the costs and social benefits, long-term ventilation was found to have become a routine and high cost intervention that conferred health benefits to most children, but without focused and continuing support to meet both the health and social needs of the child and their entire family, long-term ventilation conferred considerable social disbenefits. Methodological difficulties meant that it was not possible to enumerate the population or determine the magnitude of benefit from long-term ventilation as an intervention in different groups of children (specifically, children with profound sensory impairments). Evidence across studies demonstrated that investment in child-centred, well coordinated and effective support services could help to minimise social disbenefits and bring about the desired health and social outcomes from the perspective of the child and their family. Synthesis of findings was able to explain why parents have been successful in using Human Rights legislation to obtain previously denied resources in the absence of evidence of effectiveness.

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Acknowledgements

This thesis was completed with the support of a Research Fellowship from the Smith and Nephew Foundation and a Special Training Fellowship in Health Services Research and Health of the Public from the Medical Research Council, for which I am very grateful. I would like to thank Ian Russell, Jennie Popay and Martin Roland for guiding and supporting me through the process of applying for a MRC Fellowship.

The Smith and Nephew and Medical Research Council Fellowships have been used to develop methods in combining qualitative and quantitative methodology both in synthesising existing research and in conducting new and innovative research. Of particular importance has been the training to add a health economic dimension to the portfolio of health services research methods employed. The Fellowships provided opportunities to apply health economic methods to explore some of the very important policy issues that arose from high consumers of health services and with very vulnerable client groups. This is an under-developed area of health research and with the training undertaken as part of the Fellowships, there will be opportunities in the future to develop, extend and test methodology in this area.

The Joseph Rowntree Foundation provided additional and generous funding so that the voices of children dependent on ventilators could be included in the programme of research, and so that a secondary analysis of hospital episode statistics could be undertaken. I am particularly grateful to Emma Stone from the Joseph Rowntree Foundation for her support and for funding advisory groups to inform the research process. I am very grateful to Nick and Jenny Stephens who helped me to focus on the issues that were important to children and young people who used assisted ventilation and their families.

I am indebted to my supervisors Christine Godfrey, Ian Russell and Jennie Popay for their time, expertise and encouragement. I benefited particularly from adept feedback provided by Christine Godfrey in relation to the

preparation of the thesis for submission. I would also like to thank Jeremy Miles for commenting on a selected chapter.

I am grateful to the following people who gave advice concerning their relevant areas of expertise including: Jeni Beecham; Paul Kind, Sue Hennessey and Sue Macran from the EuroQol group; Ulrika Ravens-Sieberer from the KINDL group; John Carpenter and Janet Eyre; Michael Place and Stephen Chaplin.

I am grateful to members of the UK working party on children's long-term ventilation who acted as advisors and/or co-applicants on proposals for funding to support the empirical work and who facilitated access to the children and their families. Confidentiality prevents me from thanking individuals by name. I also am indebted to all the children, young people and their parents for warmly receiving me into their homes and providing me with such rich data to include in this thesis.

Finally, I thank Peter Finch who never once wavered in his support when I moved to a University over 100 miles from home in order to undertake a MRC Training Fellowship. I am grateful to Peter for all his help with information management, the production of this thesis, and managing two homes. Thank you also to Bryan Finch for proof reading the final draft.

Author's Declaration

All the research presented in this thesis was initiated and conducted by the author. In Chapter 2, concerning the analysis of Hospital Episode Statistics, I was the principal investigator, data were purchased from the Department of Health and statistical support was provided by Stephen Chaplin. John Carpenter and Janet Eyres provided a clinical orientation to the interpretation of findings. In Chapter 6, Jennifer Beecham provided advice and support to adapt the costing instrument.

Jane Noyes

2004

Chapter 1

The need for health services research around ventilator-dependent children and their families

1.0 Introduction

This thesis presents the findings from a programme of health services research that used and synthesised quantitative and qualitative methodologies and data to explore the epidemiology, health-related quality of life, satisfaction with services and costs of caring for children who are dependent on ventilators to survive.

Chapter 1 describes the emergence of a new population of children dependent on ventilators and provides an operational definition as to what is meant by a child dependent on a ventilator. The need for a specific health services research agenda with children dependent on ventilators is made. The moral and political context of undertaking research with children with disabilities is outlined and the impact of theories of disability on research is acknowledged. The health services research programme undertaken is outlined briefly. The introduction concludes by outlining the aims, research and methodological themes of the thesis, and summarising subsequent chapters.

1.1 The emergence of the 'ventilator-dependent' child

The National Health Service (NHS) was founded in 1948 with the expectation that overall healthcare expenditure would come down if people's health needs were met and people enjoyed good health (Foot, 1973). In contrast to this anticipated outcome, in the past 2 decades, children and adolescents have had

the highest growth rates of disability of any population cohort and the cost of their care has escalated (Laplante and Kaye, 1998; Perrin, 2002).

One such group of disabled children and young people to have emerged over the last 20 years are those dependent on ventilators to survive. It is this group of children and young people that are the focus of this thesis.

Déjà vu

This is not the first time that the United Kingdom (UK) has had to deal with the sudden emergence of a population of people requiring long-term ventilation. In the 1940s and 50s there was a worldwide epidemic of polio which rendered some patients with paralysis of the respiratory muscles (Goldberg *et al.* 1984). Large numbers of patients with polio were nursed in 'iron lungs' and considerable resources had to be diverted into caring for polio victims with respiratory failure. Polio was eradicated in the UK in the late 1950s by the development of a vaccine and mass vaccination of all children in infancy.

The modern ventilator

The modern ventilator (commonly known as a life support machine) is a technology that was originally designed for operation by highly trained medical and nursing personnel in an intensive care setting. The critically sick child who is unable to breathe unaided is attached to a ventilator in the intensive care unit via a plastic tube inserted through the nose or via a tracheostomy inserted surgically into the windpipe (trachea). The ventilator literally breathes for the child by delivering pre-programmed breaths and varying concentrations of oxygen in order to keep them alive. Constant and highly skilled nursing care is required at all times as the plastic tubes connecting the child to the ventilator can easily become disconnected leading to death or brain damage from lack of oxygen.

Recent advances in the practice of children's intensive care medicine has meant that children have survived their critical illness or accident but failed to recover the ability to breathe without the ventilator (Samuels, 1996). The

transition of the critically sick child to a ventilator-dependent child is illustrated below.

Illustration 1.1. The emergence of the child dependent on a ventilator.



Image 1.1a



Image 1.1b



Image 1.1c

Original images in colour. ©Manchester Evening News.

(1.1a). A critically sick child in the children's intensive care unit. The child is attached to a life support machine (ventilator) via plastic tubing inserted into the airway through the nose. The nurse is operating the ventilator (the white square box with controls).

(1.1b) A child who has survived their initial critical illness, but remains attached to the ventilator by a 4ft length of plastic tubing, and lives in an intensive care unit.

(1.1c) A child at home on his portable ventilator. A tracheostomy (a surgical opening into the trachea) has been formed in order to connect the child to the ventilator.

Definition

In 1998, the UK children's long-term ventilation working party defined long-term ventilation as 'any child who when medically stable, continued to need a mechanical aid for breathing which may be acknowledged after a failure to wean, 3 months after the institution of ventilation' (Jardine and Wallis, 1998).

As the incidence and prevalence of ventilator-dependent children has risen steadily, increasing numbers have survived with varying degrees of sensory and physical impairment (Noyes *et al.* 1999). The growing use of long-term ventilation as an intervention has had a considerable impact on the NHS (Davies, 1996). There is evidence that almost all children treated with long-term ventilation have consumed NHS resources by living in hospitals for months and in many cases years whilst funding has been sought to pay for them to live outside of hospital (Noyes, 1999a, 2000a, 2002). Much of this hospital stay has been taken up in expensive intensive care beds which became effectively blocked by children who no longer had a medical need to be there (Kemper, 1994; Fraser *et al.* 1997, 1998). By the early 1990s, the children's intensive care service reached a crisis point when in some units 40% of beds were blocked by children dependent on ventilators (James, 1996). A public inquiry ensued after a critically sick child died when no intensive care bed was available for him; the outcome of the inquiry resulted in the commissioning of more intensive care beds and new long-term ventilation units (Ashworth, 1996; Department of Health, 1997a,b). At the same time much debate has taken place as to whether it is in the best interests of the child to maintain their life on a ventilator, and whether the price to their family and society as a whole is worth paying (Davies, 1996; Hadden, 1996).

Over the last 10 years, long-term ventilation in children in high-income countries appears to have become a routine intervention as a consequence of circumstances, as opposed to a planned evidence based intervention. There was an obvious need to undertake research with this group of children to measure the costs and social benefits of long-term ventilation as an intervention.

1.2 The need for a health services research agenda for ventilator-dependent children

A brief overview of the literature

For a comprehensive mapping exercise and analysis of the wider grey and published literature concerning ventilator-dependent children and their families, reference can be made to a bibliography and analysis of the literature that was carried out and published at an early stage of the empirical work described in this thesis to inform the subsequent research plan (Sudbury and Noyes, 1999).

Key messages from this mapping exercise and analysis include:

- Those who have written and published about these children are mainly doctors and nurses who work in the field. This literature is made up of narrative articles, clinical material, and published guidelines (mainly based on personal practice as opposed to evidence) for the management and coordination of clinical care;
- The existing clinical literature is poor at specifying demographic features of the subjects studied and the impact of difference (race, gender, age etc);
- There is only a very small literature which presents the voices and choices of children and young people in their own way; and
- Overall, the American literature is much larger than the UK literature with Governmental concern having begun in the early 1980s as opposed to the early 1990s in the UK.

Review of the literature revealed that there had been little rigorously conducted research around children dependent on ventilators to inform decision-making. The use and type of ventilator in the intensive care setting has been subject to numerous health technology assessments to determine both efficacy and short-term outcomes for intensive care patients. In contrast there has been little research to determine the efficacy of long-term ventilation as an intervention in children and young people living in domestic settings, or how health services for

children dependent on ventilators have been managed, organised and delivered (Fulop *et al.* 2003).

A review of the few studies specifically about epidemiology, health-related quality of life and costs will be presented in subsequent relevant chapters. It is worthy of note here that very few studies around these specific issues have been undertaken with ventilator-dependent children and their families and those available are of poor methodological quality. This is not surprising as the ventilator-dependent child is a relatively new phenomenon and under-researched from a health services research perspective.

There is, in addition, a vast literature around childhood disability and arguably many of the issues around disability generally and the impact of childhood disability on families will be relevant to ventilator-dependent children. To do justice to this vast literature is beyond the scope of this thesis. Nonetheless, it is important to view children who are dependent on ventilators as part of a wider population of disabled children, many of whom are dependent on other technologies such as artificial feeding. For a review of some of the general literature on childhood disability and 'technology-dependent' children reference can be made to Sudbery and Noyes (1999).

More research has been undertaken with the adult population of ventilator-dependent people, which has expanded on a similar trajectory to the paediatric population, although for different reasons. Children with disabilities are frequently excluded from health services research because of problems concerning access and the practicalities of undertaking research with disabled and chronically ill children and their families (Perrin, 2002). This situation required addressing as the views and experiences of the children themselves may be different from those of their parents, carers or, where applicable, an adult with a similar disability. Research undertaken with, and influenced by, children and young people is more likely to address the issues that are important to them.

Research undertaken with adults cannot be applied to children as several factors distinguish children with disabilities from adults with disabilities, including: their epidemiology; the dynamic nature of children's development; children's dependence on adults for care; and the health and finance systems supporting children. These factors underpin the need, and provide an outline for a health services research agenda for children dependent on ventilators and will be explored in more detail in the following sections.

Epidemiology

As stated in the opening paragraphs of this introductory chapter, growing numbers of children and adolescents are being identified as having chronic health problems and disabilities that are different to those experienced by adults (Perrin, 2002). Health technologies such as ventilators, oxygen therapy, renal dialysis and artificial feeding have contributed to the rise in child and adolescent disability by markedly improving survival rates for many health conditions that were previously associated with higher mortality rates or incompatibility with life itself.

The long-term use of medical technologies such as ventilators has changed the nature of childhood disability in the community which is now characterised by 'high-tech' home care and increased costs to the family, NHS, social and education services (Rennick, 1995; Kirk, 1999).

The disabilities of children also have different trajectories from adults. For example, many children with congenital and rare conditions have a high mortality during childhood (such as muscular dystrophy). Whereas, some children with asthma will be disease free in adulthood.

Poor children have particularly high rates of disability, and there is strong evidence that not only does poverty increase the incidence and severity of disability but also that a child's disability may impoverish the family even more (Brunner, 1997).

Dynamic nature of children's development

Childhood disabilities need to be considered in the context of the child's stage of development because the onset, manifestations, impact, treatment and relevant outcomes of disabling conditions vary accordingly. For example, compensatory mechanisms involving movement and balance, and psychological adjustment to a spinal cord injury and dependence on a ventilator are different in babies and adolescents.

Children's dependence on adults for care

In addition to the child, research needs to take into account the child's family, their home environment and school. Children are largely dependent on their parents to provide their home and care, and to negotiate access to appropriate treatment and services. Parents' access to care, use of services, adherence to clinical regimes and ability to provide a suitable home, are determined by their health and mental health status and sociodemographic characteristics. In addition, teachers and other providers affect children's access to and the quality of services they receive in school. Parents also give their consent to treatment on behalf of their children until they reach the age of 16 years and regulate access to children for research purposes.

Health and finance systems supporting children

The various sources of public and private financing systems for the care of children with disabilities and chronic health conditions create particular problems in studying and improving the care of children dependent on ventilators, and their families. Support for children's health, social care, and education comes from a varied (and variable) mix of public, private and charitable sources. Many parents contribute a significant amount of their income to caring for their disabled child, which amounts to much more than for a non-disabled child (Dobson and Middleton, 1998). At various milestones children become eligible for specific funding streams (such as the mobility allowance at age 3), and between the ages of 16 and 18 years young people transfer into

adult funding streams to pay for their health care, social care, welfare benefits, housing allowances, continuing education etc.

The legal rights of children to express their views and opinions as service users

Health services research with children needs to take into account the views and opinions of children and young people as service users (Beresford, 1997; Ward, 1997). Children and young people are known to have different views and perspectives from those of their parents and the professionals providing their care (Morris, 1998a). Despite this, it is common that services are developed 'for' children and young people and determined by professionals and parents. The Human Rights Act (England) (1998) incorporates Articles from the United Nations (UN) Convention on the Rights of the Child and includes the legal right of children to express their views and opinions on all matters that concern them (UN, 1989).

Having established the need for a health services research agenda with children dependent on ventilators, it is important to explore briefly some contentious issues around undertaking research with people with disabilities.

1.3 Health services research with children dependent on ventilators in a moral and political context: Theorising disability

People with disabilities have campaigned for research around disability to be seen within a moral and political context and handled with great sensitivity. All research of this nature is influenced and shaped by theories of disability that can be perceived as highly contentious. Theories concerning disability powerfully influence how evidence is collected, analysed, understood and used (Alderson and Goodey, 1998a; Alderson, 2000). Widespread abuses of people with disabilities have been documented in the past, such as life-long incarceration of disabled children in closed institutions, based on a discourse

characterised by paternalism and oppression around what constituted 'in the best interests of the disabled child'.

In a contemporary health services research context, two theoretical perspectives shape research around disability: the medical model of disability and the social model of disability.

The medical model of disability

The medical model of disability views disability as a medical issue that can to varying degrees be alleviated by medical interventions that attempt to cure impairments, restore 'normal' bodily functioning and thereby improve quality of life (Marks, 1997). Outcomes are measured by comparing mortality and functional status (morbidity) of disabled people against normative data derived from 'normal' healthy people within society.

The social model of disability

In contrast, the social model of disability has been developed by people who themselves have disabilities and whom to varying degrees reject the dominant discourse of medical paternalism and the discourse around what constitutes 'normality' (Marks, 1997). The social model conceptualises disability as a social and political issue rather than a medical one. Whilst respecting the value of scientifically based medical research, the social model of disability calls for social and political solutions to change disabling discrimination in order to improve the quality of disabled people's lives. This has involved redefining what is meant by disability, with a clear distinction being made between impairment and disability. Impairment is defined as being the functional limitation within the individual caused by physical, mental or sensory impairment. Disability is the loss or limitation of opportunities to take part in the ordinary life of the community on an equal level with others, because of physical or social barriers (Oliver, 1998).

In this thesis both the medical and social model of disability are used to underpin the empirical work, and methodological work has been undertaken to

compare, contrast and synthesise findings that look at similar research questions from different methodological and theoretical perspectives.

1.4 Operationalising a health services research agenda with children dependent on ventilators

Ensuring that children who are dependent on ventilators have a good quality of life within the available resources, presents both NHS and social care providers with numerous challenges, including the need to:

- Commission services based on actual and projected numbers of children dependent on ventilators;
- Provide services that have a positive impact on the quality of life of children and their families;
- Deliver services that are cost-effective, and
- Involve children and carers to ensure that services are planned around the child and family, and meet their identified needs.

Undertaking research with this emerging population of children and their families had been particularly difficult, due to issues around locating and accessing children who were distributed throughout the UK. Access to ventilator-dependent children was made easier in 1996, when a group of doctors, nurses, social workers and various therapists involved in the care of children dependent on ventilators formed the UK working party on children's long-term ventilation. One of the aims of the group was to undertake research and to facilitate access to combined patient caseloads to ensure sufficient numbers of children were included in research studies.

A period of exploratory fieldwork, an initial exploratory qualitative study (Noyes, 1999a) and a review the existing literature (Sudbury and Noyes, 1999) was undertaken; findings were used subsequently to frame a comprehensive health services research agenda around children dependent on ventilators. This thesis is part of a wider body of research to fulfil this agenda that was supported by a Medical Research Council Fellowship in Health Services Research.

Themes of the thesis

This thesis is focused on four research themes:

1. The origins and size of the population;
2. The health-related quality of life of children;
3. The costs of caring, and
4. Satisfaction with services.

Cross cutting these themes are a range of methodological challenges.

Methodological sub-theme

The thesis will explore the application and synthesis of quantitative and qualitative methodologies as a broad methodological sub-theme. For each of the four major research themes outlined above, quantitative and qualitative methods will be applied to address specific research questions that were developed and shaped following a period of exploratory work and review of the literature. The synthesis of findings from studies embedded in quantitative and qualitative paradigms will be used to build up an overall picture of the costs and social benefits of long-term ventilation as an intervention in children.

Thesis outline

Chapter 2 focuses on the origins and size of the population (theme 1). In order to establish a context for this thesis, current evidence concerning estimates of incidence and prevalence of ventilator-dependency in childhood is reviewed. Findings from quantitative and qualitative epidemiological approaches to health needs assessment are then reported. The quantitative study aims to determine whether the population and resource use of ventilator-dependent children can be enumerated from the secondary analysis of routinely collected NHS data. Whereas, the qualitative study explores with healthcare professionals the reasons and factors that have contributed to the emergence of this new population. The chapter concludes by synthesising findings across studies.

Chapters 3, 4 and 5 are linked sequentially and focus on the health-related quality of life of ventilator-dependent children (theme 2).

Chapter 3 begins by creating a context concerning the conceptualisation of health and quality of life by reviewing briefly the literature around the measurement of the quality and quantity of a child's life. In order to explain the state of current practice around long-term ventilation as an intervention, legal test cases around withdrawing and withholding treatment and the impact of the Human Rights Act (1998) on decision-making are discussed. The chapter then reports a quantitative study underpinned by the medical model of disability that aims to measure health-related quality of life in 35 ventilator-dependent children and young people. This quantitative study aimed to address whether ventilator-dependent children had lower health-related quality of life than a normative population of school children; whether ventilator-dependent children's self-reports of their health-related quality of life differed from those of their parents; and whether the health-related quality of life of ventilator-dependent children living in hospital differed from those children living at home.

Chapter 4 continues to focus on the health-related quality of life of children (theme 2) by reporting a qualitative study underpinned by the social model of disability that aims to define and describe health-related quality life of children dependent on ventilators from the perspective of the children themselves and their parents. Findings are then framed around the Articles of the Human Rights Act, which has been used subsequently by parents to secure resources previously denied them through other routes.

In **Chapter 5** a synthesis of quantitative and qualitative findings from the empirical studies concerning the health-related quality of life of ventilator-dependent children in Chapters 3 and 4 is presented (theme 2).

Chapters 6, 7 and 8 are linked sequentially and focus on the costs of caring for children dependent on ventilators (theme 3) and satisfaction with services (theme 4).

In **Chapter 6** the costs of care are first explored through legal imperatives around the allocation of finite resources. The chapter then reports an economic study underpinned by the medical model of disability, that aims to estimate the long-run marginal service costs of supporting ventilator-dependent children in hospital, home and residential settings over 1 year; levels of satisfaction with services received are also measured and gaps in service provision estimated.

Chapter 7 continues the focus on the costs of care (theme 3) and satisfaction with services (theme 4) by describing a qualitative study underpinned by the social model of disability that aims to explore the financial impacts to families of caring for ventilator-dependent children, and describe how satisfied parents and children are with the services they received.

In **Chapter 8**, a synthesis of quantitative and qualitative findings from the empirical studies concerning the costs of care (theme 3) and satisfaction with services (theme 4) in Chapters 6 and 7 is presented.

Chapter 9 has three distinct parts. First, a synthesis of findings derived from the 4 research themes concerning the costs and benefits of long-term ventilation in children is presented. Findings are assessed against a set of criteria for framing evidence around health problems and medical conditions. The contribution of findings to the evidence base is summarised and remaining gaps highlighted. The second section discusses in further detail one of the major conceptual issues to emerge from the synthesis of findings concerning the lack of clarity regarding the needs of ventilator-dependent children and outcomes of services provided. Finally, synthesised findings are reviewed in the light of current health and social policy initiatives.

The thesis concludes with a final chapter (**Chapter 10**) that outlines future research directions and makes the case for a more extensive evidence base concerning small volume high costs groups such as ventilator-dependent children. The thesis closes by exploring the unresolved dilemma as to what level of resources, and which health and social outcomes, the state should reasonably be expected to meet.

Chapter 2

Enumerating the population and describing the emergence of children and young people dependent on ventilators in the UK

2.0 Introduction

Chapter 1 chronicled the emergence of a new a population of children who survived critical illnesses and accidents but remained dependent on ventilators and who spent prolonged periods of time in hospital.

Chapter 2 focuses on the origins and size of the population of children dependent on ventilators in the UK (theme 1). In this chapter, quantitative methods are used in an attempt to enumerate the size of the population and qualitative methods are used to explore and explain the factors and reasons that have contributed to the emergence of this population.

The chapter begins by exploring epidemiological approaches to health needs assessment and presents a critical review of surveys that have been undertaken to enumerate the size of the population and describe the underlying diagnoses of the children and young people.

The chapter then goes on to describe a secondary analysis of NHS hospital episode statistics concerning hospital discharges in England and Scotland to map the numbers, characteristics and in-patient bed use of children who spent prolonged periods of time in hospital, and more specifically to ascertain whether children dependent on ventilators could be identified from routinely collected data using International Classification of Disease (ICD) coding. Strengths and weaknesses of the method used are highlighted.

Finally, qualitative findings derived from fieldwork undertaken with healthcare professionals exploring the reasons and factors that have contributed to the emergence of this new population are described. Strengths and weaknesses of data derived from exploratory fieldwork are outlined. The chapter concludes by synthesising findings across studies.

2.1 Epidemiological approaches to health needs assessment

Epidemiology is concerned with the study of the distribution and determinants of health-related states or events in specific populations (Wright *et al.* 1998). Williams and Wright (1998) define descriptive epidemiology as a method that describes the occurrence of disease in terms of person, place and time:

- Person – who the affected people are (in terms of their age, sex, occupation, socioeconomic group, etc);
- Place – where and when they get diseases and in what way prevalence and incidence vary geographically (locally, regionally, nationally, or internationally), and
- Time – when people get diseases, whether this varies by, for example, season; and how disease occurrence is changing over time.

Incidence is the number of new cases observed and prevalence is the number of cases present in the population. Incidence is a rate that is calculated over a specified period of time and prevalence is a proportion that is measured at a point in time, but does not have a time dimension.

Epidemiology is framed by the medical model of health need, viewing need in terms of the occurrence of specific diseases and health-related states. Epidemiological methods can be used to describe health need in terms of the distribution of specific health states and diseases; although incidence and prevalence do not necessarily equate with need, they are important in describing the population burden of the disease or health state. Epidemiological data can be applied subsequently to consider the ways in

which services are delivered and the effectiveness and cost-effectiveness of interventions to meet identified need.

Planning service provision for children dependent on ventilators

Commissioners of services for children dependent on ventilators require epidemiological data to implement policies and plan services. Studying incidence and prevalence of ventilator-dependency in an entire population, however, is not straightforward and presents a number of methodological challenges. Primarily, ventilator-dependency is not a disease, rather it is an iatrogenic intervention; seriously ill children and young people with a multitude of diseases and disease states are placed on ventilators by doctors. The majority of children recover sufficiently to breathe on their own. A small and as yet unknown proportion of children and young people remain dependent on the ventilator and require long-term care. The only thing that these children have in common is their reliance on the same type of medical device – the ventilator. Some, but not all, children with similar disease states require assisted ventilation so it is not possible to identify all children with a particular diagnosis and assume they are dependent on a ventilator. There is also variation in the severity of ventilator-dependency from children with complete failure of respiration who require artificial ventilation 24 hours a day and would die if disconnected, to children with mild failure of respiration who only require artificial ventilation to supplement their own breathing.

2.2 The need to enumerate the size of the population

During exploratory fieldwork it was reported by commissioners of services that they had never envisaged (or planned) that such large sums of money would be required to support so many disabled children with increasingly complex needs for health and social care to live at home. In order to provide appropriate provision of services for children dependent on ventilators, commissioners of health and social services said that they needed to have accurate information concerning incidence, prevalence and resource use.

The need for accurate epidemiological estimates is especially pressing as, following a recent judicial review, the Department of Health conceded that money should be made available for a child dependent on a ventilator to be cared for at home. The plaintiff argued that under the Human Rights Act (1998) the child's right to a family life and privacy was being violated by her prolonged stay in hospital (*N v Department of Health in Northern Ireland*, 2003).

The outcome of this case will have benefits for all children who are dependent on ventilators as they can use this case to enforce their legal right for resources to live at home. On the other hand, fieldwork interviews with two commissioners of services revealed their anxiety that this judgment was equivalent to them signing a blank cheque as they had no robust information concerning the numbers of children involved and limited resources available to them to provide services.

2.3 What is currently known about the incidence and prevalence of children dependent on ventilators in the UK?

There are currently two national and one regional surveys that attempt to identify the incidence and prevalence of ventilator-dependent children in the UK. One survey (Robinson, 1990) was published prior to the commencement of empirical work reported in this thesis; the other two (Jardine *et al.* 1999 and Margolan, 2001) were published during the study period.

Robinson (1990) surveyed all paediatricians in the UK at 2 time points in 1983 and again in 1987. He defined ventilator-dependent children as those being on 'total ventilator support' for 6 months or more, but excluded children who used non-invasive methods of ventilation (such as ventilation delivered via a face mask).

Of the 35 children Robinson identified, 10 were ventilated mainly at home and cared for by their families. An additional 5 children mainly lived in hospital and spent short periods of time at home. The remaining 20 children had been

managed entirely in hospital and lived principally in intensive care units. Numbers in broad diagnostic categories with sex distribution and age range are set out in Table 2.1. Nine children left the register during the 2 sampling periods (1983 and 1987). One had left the paediatric age range, and 2 had been successfully weaned from the ventilator; 6 had died.

Table 2.1. Diagnosis and numbers of children, sex and age range (1983-1988). Robinson (1990).

Diagnosis	No	Boys	Girls
High cervical trauma	14	9	5
Neuromuscular disease	7	2	5
Primary nocturnal hypoventilation	6	3	3
Infection	3	2	1
Tumour	2	1	1
Bronchopulmonary dysplasia	2	2	0
Vascular malformation	1	0	1

Incidence and prevalence of children notified to the study by calendar year are set out in Table 2.2.

Table 2.2. Incidence and prevalence of ventilator-dependent children by calendar year (1975-88) in the United Kingdom*. Robinson (1990).

Year	75	76	77	78	79	80	81	82	83	84	85	86	87	88
Incidence	1	0	1	1	1	2	2	2	4	4	2	10	3	0
Prevalence	1	1	2	3	4	6	8	10	14	17	17	24	24	24

* Data are missing on 2 children

The geographical distribution of children is shown in Figure 2.1. overleaf.

Figure 2.1. Geographical distribution of ventilator-dependent children in the UK 1988. Robinson (1990).



Robinson's questionnaire survey has a number of methodological flaws. Case finding was dependent on voluntary reporting by a single source (paediatricians) and an upper age limit for childhood was not given. Data were collected retrospectively and it is probable that numbers of children were ventilated previously that had either died or were weaned from the ventilator before the survey was conducted. Back in 1988, a universal definition for 'ventilator-dependency' had not yet been agreed so Robinson came up with his own definition but excluded children who used non-invasive ventilation even if they were 'dependent' on a ventilator for more than 6 months. This is an inconsistency and introduced an unacceptable level of bias due to under reporting of legitimate cases.

Despite these flaws, Robinson provided the first attempt to establish the incidence and prevalence of this new group of children and confirmed that 20 children were living inappropriately in intensive care units and thereby blocking beds.

A national crisis: Blocking of children's intensive care beds

During the 1990s, paediatricians also reported that increasing numbers of children dependent on ventilators were living inappropriately in children's intensive care units and blocking this scarce resource for other critically sick children (James, 1996; Fraser *et al.* 1997). Doctors appeared powerless to manage the situation effectively as it was not considered common or safe practice to transfer these children to children's wards or, for that matter, home. The services for these children to be cared for safely at home had not yet been established and only those children with very motivated and determined parents were discharged home (Noyes, 1999a, 2002).

By 1996, 42% of intensive care beds at Great Ormond Street hospital for children were blocked by children dependent on ventilators resulting in the refusal of 276 children who were referred for intensive care during 1995 (James, 1996). This pattern was found to be repeated throughout children's intensive care units in the UK (Fraser *et al.* 1997).

The issue of blocked intensive care beds became a national issue later in 1996 when a boy from Manchester died, as an intensive care bed was not available for him due to blocked beds. A public inquiry followed and a National Coordinating Group for paediatric intensive care was set up by the then Conservative Government (Ashworth, 1996). The subsequent report (Department of Health, 1997,a,b) called for a second national audit of the numbers of children requiring long-term ventilation and for regional offices of the NHS Executive and health authorities to 'assess the requirements of children who need long-term ventilation'.

Jardine *et al.* (1999) in association with the members of the UK working party on children's long-term ventilation (of which the author is a member) were commissioned by the Department of Health to estimate the incidence, prevalence, underlying diagnosis, ventilatory needs and location of children dependent on ventilators in the UK. A postal questionnaire was sent to 112

consultant respiratory paediatricians and 49 lead clinicians of paediatric and neonatal intensive care in the United Kingdom.

For the purposes of this survey, members of the UK working group defined long-term ventilation as:

‘Any child who when medically stable, continued to need a mechanical aid for breathing which may be acknowledged after a failure to wean, 3 months after the institution of ventilation’.

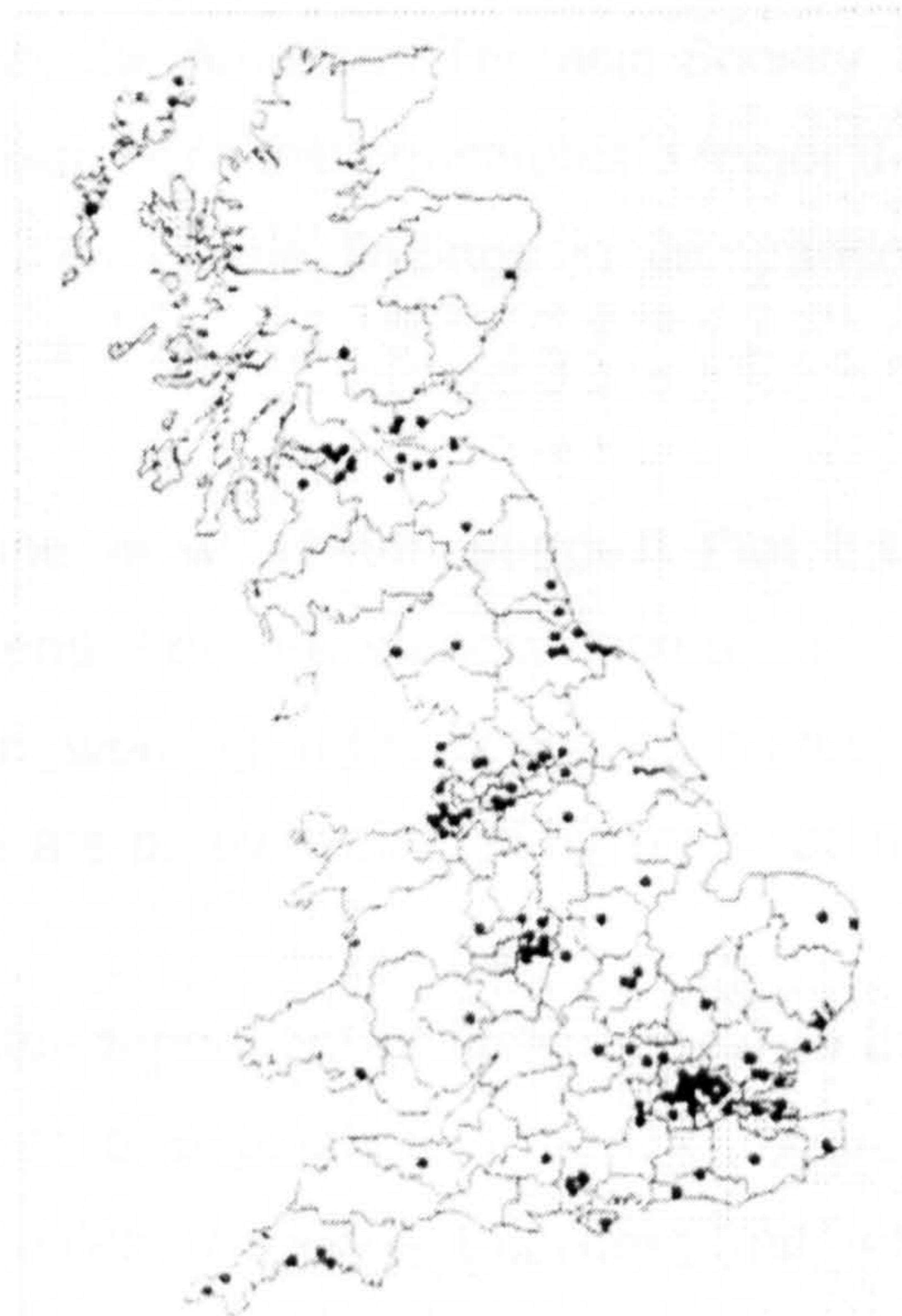
Each child was identified by name, date of birth and postcode to avoid duplication of cases. Parental consent was required before the doctor returned the questionnaire. Eighty-nine consultant respiratory paediatricians responded (80%) and 30 lead clinicians from paediatric and neonatal intensive care (61%). The survey identified 141 children and detailed information was obtained on 136 children. The main diagnostic categories and location of the children are described in Table 2.3. These are not entirely consistent with the diagnostic categories used by Robinson (1990) which makes comparison difficult. Two diagnostic categories are identical and can be compared directly; neuromuscular disease and bronchopulmonary dysplasia. Robinson reported 7 children with neuromuscular disease, whereas Jardine *et al.* identified 62. This probably reflects the increasing numbers of children such as boys with muscular dystrophy who are now ventilated at the end of life as their respiratory function fails; in the past these boys would have died. For the second category bronchopulmonary dysplasia (lung disease in premature infants), Robinson reported 2 cases, whereas Jardine *et al.* identified 6. This supports findings from fieldwork interviews with doctors who said that in their clinical experience the introduction of the pharmaceutical product surfactant increased the survival of premature infants but with increased morbidity.

Table 2.3. Diagnoses of children dependent on ventilators identified by Jardine *et al.* (1999).

Disorder	No at home	No in hospital
Neuromuscular disease	52	10
Congenital central hypoventilation syndrome	13	5
Spinal injury	10	6
Craniofacial syndrome	9	0
Bronchopulmonary dysplasia	1	5
Other	8	17
Total	93	43

Of the 136 children, 71 were receiving non-invasive ventilation, which underlines the weakness of Robinson's earlier survey that failed to include them. Over two thirds of the children were at home, which is in contrast to the previous survey where two thirds of the children were in hospital. Geographical distribution is illustrated in Figure 2.2 and indicates an increase in density since Robinson's earlier survey – especially in urban areas.

Figure 2.2. Geographical distribution of 136 ventilator-dependent children (Jardine *et al.* 1999).



Jardine *et al.* (1999) demonstrated a consistent increase in incidence and prevalence (Table 2.4) which correlates with the experience of paediatric intensive care doctors that the beds in their units were becoming blocked by this group of children from the early 1990s onwards (James, 1996; Fraser *et al.* 1997).

Table 2.4. Incidence of children in the United Kingdom dependent on ventilatory support by calendar year 1988-1997. Jardine *et al.* (1999).

Year	88	89	90	91	92	93	94	95	96	97*	Total
Incidence	1	2	4	4	11	19	11	27	28	18	125

* until September 1997

Jardine *et al.*'s survey has a number of methodological weaknesses. Jardine (along with the UK LTV group) developed their own consensus definition for ventilator-dependency. This definition appears to be open to interpretation and lacking in precision. The researchers acknowledge that issues such as 'medically stable' had to be left to the judgement of the child's consultant and as such may be subject to considerable bias. This definition also differs slightly from the one agreed by the American Thoracic Society (1990). Not having an internationally recognised definition constitutes a major threat to external validity and makes interpretation of the findings in comparison to the international literature difficult.

Estimates from Jardine *et al.* (1999) suggest that the numbers of children requiring long-term ventilation are steadily increasing. This is in line with the experiences of doctors working in the speciality, although it is difficult to know whether the increases are partly explained by improved notification.

At first glance ventilator-dependent children appear to be clustered around the major cities and centres of population in the UK (Figure 2.2). There are areas with apparently no ventilator-dependent children and yet during early fieldwork for the empirical work reported in this thesis, 60 cases were identified by cascade sampling, many of whom were later found not to be entered onto the database (Noyes, 1999a).

Jardine *et al.* report incidence rates by calendar year (Table 2.4). These figures do not include young people over the age of 17 years who may have become dependent on ventilation during the years 1988-1997. Neither were mortality figures collected for ventilator-dependent children who died during this period.

Like Robinson, Jardine *et al.* used a single source (doctors) to identify cases and this is a major threat to the validity of the survey. A number of validated methods to estimate small and hard to reach populations could have been used. For example, Sudman *et al.* (1988) suggest that 2 sources should be used to identify the children – such as medical settings and educational settings, and validated modelling techniques can be used to estimate the number of children not detected from either medical settings or schools.

Jardine *et al.* (1999) reported difficulty in obtaining data from some centres. If resources had been available it may have been possible to divide the UK into manageable units and allocate a local researcher to find cases using Sudman *et al.*'s method.

Following publication of the national survey in the British Medical Journal, the specialist commissioning group in the South West of England published their own local survey (Margolan, 2001; Lenton *et al.* 2001). Lenton *et al.* (2001) designed a cross sectional point prevalence survey to ascertain the numbers of children requiring long-term ventilation in the South West of England during November 1999.

A questionnaire was sent to all clinical directors of paediatric intensive care in the South West region (defined as the region as managed by the NHS executive in the South West of England). The questionnaire asked consultants to identify children either in hospital or in the community who were currently ventilated for greater than 3 months (in line with Jardine's definition). The parents of children who were identified were invited to participate in a qualitative interview and amongst other things were asked to identify any other children in the region.

Ten out of 11 (91%) clinical directors responded. Eighteen children were identified (3 in hospital, 15 at home and 2 had died in hospital) giving a prevalence of 1.63/100,000 in the 0-19 year old population.

This survey used 2 sources to identify cases (clinical directors and parents of children identified), which strengthen the internal validity of the study. Clinical directors in the South West received their funding from the specialist commissioning group and were in regular contact with the researchers. It was felt that they would be able to most reliably identify these children and had a vested interest to do so. Parents demonstrated that they had knowledge of children other than their own, but no additional cases were identified. There is a danger that children may be managed by doctors in other parts of the service and that parents live in isolation to other cases, but despite these weaknesses, this regional survey is likely to be the most accurate regional point prevalence survey available to date.

Comparison of UK incidence and prevalence with international data

Surveys using similar methodologies and a single source for case reporting were undertaken in Canada and Switzerland at a similar point in time to Jardine *et al.* (1999) and identified numbers of children comparable per head of population (Dhillon *et al.* 1996; Kamm *et al.* 2001). In contrast, in France where a greater proportion of Gross Domestic Product is spent on state-funded healthcare, and reporting of new cases is more advanced than in the UK; incidence rates appear higher with 158 children recorded as being commenced on long-term ventilation at home during 12 months in 1992 (Fauroux *et al.* 1994). Fauroux *et al.* do not however state the prevalence of ventilator-dependent children in the general population. A recent single source survey in Japan identified 434 children dependent on ventilators for more than 3 months; these high numbers were attributed to parental decision-making and availability of free healthcare for these children, most of who lived in hospitals (Sakakihara *et al.* 1996).

Country-wide surveys have not been undertaken in the United States and state-wide point prevalence surveys have been hampered by the disparate nature of healthcare providers and provision of services. One exception was Palfrey *et al.* (1991) who carried out a state-wide census in Massachusetts in 1987 using a capture-recapture technique. They identified 1244 technology-dependent children (0.08% of the total state child population), 41 of whom were ventilator-dependent children. There is also evidence from the U.S. Office of Technology Assessment that increases in incidence and prevalence of ventilator-dependent children began to put pressure on the availability of service provision from the mid 1980s onwards, over a decade earlier than in Europe (US Office of Technology Assessment, 1987, US Department of Health and Human Services, 1988).

2.4 What else can be done to estimate the size of the problem?

As all cases of long-term ventilation are initiated in hospital and it is known that this group of children have frequent in-patient episodes, it was decided to determine whether the population and resource use of ventilator-dependent children and young people could be enumerated from routinely collected NHS data.

National Hospital Datasets

In order to take this work forward, it was decided to form a collaboration with colleagues from the York Health Economic Consortium who were experienced at working with large datasets of this nature. It was decided subsequently that the next stage would be to establish how much of the desired information could be obtained from the respective national hospital data sets: HES (Hospital Episode Statistics) data for England and SMR (Scottish Morbidity Record) data for Scotland.

Both the HES (England) and SMR (Scotland) databases contained personal, medical and administrative details of all patients admitted to and treated in NHS

hospitals. HES data covers all NHS Trusts in England; private hospitals are not covered, although it does include private patients who were treated in NHS hospitals.

Scottish Morbidity Record data is Scotland's equivalent data to hospital episode statistics. A SMR record is generated for patients receiving care in general/acute specialties.

Both the English and Scottish datasets are coded with the International Statistical Classification of Diseases and Related Health Problems -10th Revision (DoH, 2002d). This classification system is abbreviated to ICD-10 and codes are organised into groups called chapters. Codes consist of a single letter followed by 3 or more digits. The diagnoses are presented in code order (rather than by the diagnosis name). The database administrators reported that ventilator-dependency was recorded within the ICD chapter of codes Z00 – Z99: Factors influencing health status and contact with health services. Within this chapter, the relevant codes for a patient dependent on a respirator (Z99.1) and for a patient dependent on another machine (Z99.8) are found. Each NHS patient is ascribed a unique identifier (NHS number) which should be entered by coders for each completed hospital episode. The NHS number is constant whereas patients are ascribed different patient numbers by different NHS facilities.

Consultation with the database administrators confirmed that a secondary analysis of hospital episode statistics focusing on children who experience prolonged admissions and or ventilator-dependency in hospital had not been undertaken before.

Methods

A secondary analysis of routinely collected HES and SMR records was planned, modified according to available resources and undertaken. The author obtained the funding, refined the research questions, prepared a specification and submitted it to the respective database administrators for approval and costing.

Originally a proposal was submitted to analyse all hospital discharge data for children age 0-19 years for the years 1995-1999, but unexpectedly this turned out to be prohibitively expensive (costing 6 times any previous analysis undertaken by the York Health Economic Consortium and beyond the allocated budget). After consultation with the advisory group and funder, it was decided to scale down the proposal and purchase data to look at one year 1999-2000 (the most recently released) and limit the number of diagnoses to the primary diagnoses (ICD coders can record up to 3 per child).

During exploratory fieldwork, it was also established that children dependent on ventilators had the most prolonged admissions in hospital after initial diagnosis; almost always greater than 6 months. As the vast majority of children spend less than 2 days in hospital in one admission it was decided to limit initially the analysis to finished consultant episodes lasting 4 weeks or more (thereby excluding around 90% of short routine admissions and making the purchase of data affordable). A revised proposal was submitted to the database administrators requesting discharge data covering all patients aged <20 years at admission, who spent one month or more in one spell in NHS hospitals in England and Scotland for the year 1999-2000.

Emerging data were presented to the Project Advisory Group for scrutiny. Professor John Carpenter from the University of Durham and Professor Janet Eyre from the University of Newcastle provided a clinical focus to the interpretation of findings. Queries were referred back to the relevant authorities for clarification. Stephen Chaplin from the York Health Economics Consortium provided technical support with the data analysis.

Aims and objectives

The study had 2 aims:

1. To map the numbers and characteristics of children and young people aged <20 years at admission, who were discharged from NHS hospitals in England and Scotland between 1st April 1999 and 31st March 2000 having spent one month or more in one in-patient spell, and

2. To see if it was possible to identify children and young people who were dependent on ventilators from routine ICD coding.

The objectives were to identify:

- Total numbers of children and young people;
- Their age, sex and ethnicity;
- Their primary diagnosis ascribed at discharge; and to look specifically for children dependent on ventilators, and
- How long they stayed in hospital.

Data analysis

Both datasets were provided on compact disc. The English HES data arrived in the form of pre-specified tables in Excel™ (Microsoft, 2000). The Scottish SMR database administrator sent pre-specified raw data in Excel™ for the team to analyse.

Results

How many children?

In England, there were 16,321 children and young people with a length of stay greater than 1 month (Table 2.5). Over half of these (9,149 - 56%) were babies of whom 62% had been born in hospital. The majority (13,486 - 83%) were in hospital for between 1 and 2 months. Over 900 children and young people spent over 6 months in hospital and a third of these (333) were there for 12 months or longer.

Table 2.5. Discharge data from NHS Hospitals in England 1999-2000. Age at admission by length of stay.

	Age in years at Admission					Total	%
	<1	1-6	7-12	13-16	17-19		
Length of stay (months)							
1-2	8161	1250	1095	1174	1806	13486	82.6%
3-5	860	105	157	323	477	1922	11.8%
6-8	68	34	43	114	157	416	2.6%
9-11	16	9	18	62	59	164	1.0%
12+	44	55	48	86	100	333	2.0%
Total	9149	1453	1361	1759	2599	16321	
%	56.1%	8.9%	8.3%	10.8%	15.9%		100%

In Scotland, like England, the majority of children stayed between 1 and 2 months (64%). Over half were younger than 6 years old at admission (Table 2.6). Twenty-one individuals stayed in hospital for longer than 12 months; 1.5 children in a hundred, which was a slightly lower rate than in England (2 per hundred).

Because inadvertently the SMR administrator sent discharge data for all children aged 0-19 years in Scotland, including those that stayed less than 1 month, it was possible to confirm that the average length of stay across all admissions was just under 2 days.

Table 2.6. Discharge data from NHS Hospitals in Scotland 1999-2000. Age at admission by length of stay.

	Age in years at admission					Total	%
	<1	1-6	7-12	13-16	17-19		
Length of Stay (months)							
1-2	444	125	91	93	139	892	63.7
3-5	171	27	27	53	109	387	27.7
6-8	19	5	7	20	20	71	5.1
9-11	4	2	3	6	13	28	2.0
12+	1	6	3	2	9	21	1.5
Total	639	165	131	174	290	1399	
%	45.7%	11.8%	9.4%	12.4%	20.7%		100%

Sex ratio

In both countries the largest proportion of individuals were male (Table 2.7).

Table 2.7. Discharge data from NHS Hospitals 1999-2000. Children's/young person's sex by area.

Sex	England		Scotland	
	No	%	No	%
Male	8831	54.3%	794	56.8%
Female	7439	45.7%	605	43.2%
Total	16270	100%	1399	100%

Ethnic group

In England, 40% of the patients had no information concerning ethnicity recorded. It was not possible to draw any conclusions from this data. In Scotland, data on ethnicity was not recorded.

Why were the children and young people in hospital?

Table 2.8 shows the top 10 ICD-10 codes ascribed to children and young people in England at discharge. The top 10 ICD-10 codes represent 93% of all in-patient admissions, easily the largest proportion of these admissions were for conditions arising shortly after birth (6,699 - 42%), which reflects the large proportion of babies as highlighted in Table 2.5.

3.5 % (571) were ascribed to ICD group Z00 - Z99: Factors influencing health status and contact with health services. These categories are provided for occasions when circumstances other than a disease or injury are recorded as diagnoses or problems. Codes Z80-Z99 relate to conditions that influence health status and within this group Z99 provides codes for dependence on an enabling machine. Code Z99.1 is for dependence on a ventilator and Z99.8 is for dependence on other enabling machines and devices. It was not possible to determine individual Z codes from the pre-formatted tables provided.

Table 2.8. Discharge data from NHS Hospitals in England 1999-2000. Top 10 ICD-10 codes by age.

ICD-10 Descriptor	Age in years at admission					Total	%
	<1	1-6	7-12	13-16	17-19		
P00 - P96: Certain conditions originating in the perinatal period	6695	4	0	0	0	6699	41.7
F00 - F99: Mental and behavioural disorders	3	28	234	682	1286	2233	13.9
S00 - T98: Injury, poisoning and certain other consequences of external causes	58	411	371	216	272	1328	8.3
R00 - R99: Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	509	131	116	195	340	1291	8.0
Q00 - Q99: Congenital malformations, deformations and chromosomal abnormalities	751	113	51	33	14	962	6.0
C00 - D48: Neoplasms	53	205	148	85	103	594	3.7
Z00 - Z99: Factors influencing health status and contact with health services	334	46	38	84	69	571	3.5
G00 - G99: Diseases of the nervous system	77	91	93	116	85	462	2.9
K00 - K93: Diseases of the digestive system	106	73	56	65	66	366	2.3
J00 - J99: Diseases of the respiratory system	155	110	35	30	33	363	2.3
Other	228	238	213	235	267	1181	7.4
Total	8969	1450	1355	1741	2535	16050	
%	55.9%	9.0%	8.4%	10.9%	15.8%		100%

The top 10 ICD-10 codes in Scotland (Table 2.9) represent 94% of total admissions. Like England, the 2 largest groups were for conditions arising shortly after birth (30%), and mental and behavioural disorders (23%). 3.1% (43) were ascribed to ICD group Z00 - Z99: Factors influencing health status and contact with health services. As the raw data was provided, it was possible to confirm that ICD codes Z99.1 and Z99.8 were not ascribed as a primary diagnosis or health problem (secondary diagnostic codes were not provided). It would seem likely from this evidence that there is no simple means of identifying ventilator dependent children from primary diagnostic ICD codes.

Table 2.9. Discharge data from NHS hospitals in Scotland 1999-2000. Top 10 ICD-10 codes by age.

ICD-10 code	Age in years at admission					Total	%
	<1	1-6	7-12	13-16	17-19		
P01-P96: Certain conditions originating in the perinatal period	419	0	0	0	0	419	30
F00-F99: Mental and behavioural disorders		1	27	89	203	319	22.8
S00-T98: Injury, poisoning and certain other consequences of external causes	2	48	40	19	21	130	9.3
Q00-Q99: Congenital malformations, deformations and chromosomal abnormalities	105	14	2			121	8.6
C00-D48: Neoplasms	10	36	19	15	8	88	6.3
G00-G99: Diseases of the nervous system	17	10	7	8	6	48	3.4
K00-K93: Diseases of the digestive system	12	9	9	6	10	46	3.3
J00-J99: Diseases of the respiratory system	14	9	4	2	4	33	2.4
Z00-Z99: Factors influencing health status and contact with health services	6	6	4	10	17	43	3.1
R00-R99: Symptoms, signs and abnormal clinical and laboratory findings, not elsewhere classified	10	6	5	4	2	27	1.9
A00-B99: Certain infectious and parasitic diseases	7	8	4	1		20	1.4
E00-E90: Endocrine, nutritional and metabolic diseases	7	5	2	3	2	19	1.4
Other	30	14	8	17	17	86	6.1
Total	639	166	131	174	290	1399	
%	45.7%	11.8%	9.4%	12.4%	20.7%	100%	100%

How long were the children and young people in hospital?

Average length of stay comparisons could not be made directly between England and Scotland because of the way the data was collected. There are occasions when the HES system might not generate a Finished Consultant Episode but the Scottish data does, for example, when a patient changes specialty but remains with the same consultant. To make these data more comparable continuous in-patient stays were calculated for the Scottish data by linking medical records for individual patients. The average length of stay for those who were in hospital longer than 12 months was 3889 days or 10.6* years (England) and 1056 days or 2.9 years (Scotland).

* Length of stay data for England contain methodological flaws and should not be quoted out of context (see discussion below).

It was not possible to make direct comparisons between England and Scotland regarding the average length of stay because of the ways the data were

collected. However, there was some suggestion that the average length of stay across all groups was longer in England, 141 days versus 82 days in Scotland. This overall difference could be largely accounted for by those children and young people who had been in hospital longer than 12 months; the average length of stay was around 10.6 years in England and nearly 3 years in Scotland. The long average length of stay in England warranted further exploration as it did not appear to match the experience of the paediatrician advising the study who said that 10.6 years appeared too long. These data were checked with the HES project team who confirmed that they were as received from the NHS Trusts. An interim report was submitted to the Department of Health for scrutiny and advice. Although there was no more funding available, the Department of Health agreed to release further data to help explain the findings.

Breakdown of length of stays of children and young people staying over 12 months in NHS Hospitals in England

A breakdown of length of stay for this group of children in England is presented in Table 2.10. Of this group, 178 (53%) stayed for 1-2 years; 223 (67%) stayed for between 1-4 years. This sub-set of children and young people may be contrasted with 77 people (23%) who were recorded as having spent between 25 and 60 years in the hospital from which they had been discharged, and one person over 60 years. Clearly, these 2 subgroups are very different; the group of 77, for example, were obviously no longer children but adults. Many of these appear to be adults who were admitted as children to long-stay learning disability and psychiatric hospitals and, very many years later, discharged to 'care in the community'. This provided an explanation as to why the average length of stay was so high for this group. In Scotland, this group of adult patients were not included in the paediatric dataset.

The paediatrician advising the study confirmed that his NHS Trust had discharged 2 ventilator-dependent children who had spent 1.8 and 2.5 years respectively in hospital during the census period. These children will have been included in this group of 333 individuals who were discharged following an admission of more than 12 months, but it was not possible to identify them with the methods used in the current study.

Table 2.10. Discharge data from NHS hospitals in England 1999-2000. Distribution of length of stay for all children/young people aged under 20 when admitted and staying more than 12 months.

Length of stay	Number	%
12-13 months	84	25.2
13-14 months	18	5.4
14-15 months	17	5.1
15-18 months	33	9.9
18-21 months	14	4.2
21-24 months	12	3.6
2-2.5 years	22	6.6
2.5-3 years	9	2.7
3-4 years	14	4.2
4-5 years	2	0.6
5-6 years	2	0.6
6-7 years	8	2.4
7-8 years	0	0.0
8-9 years	2	0.6
9-10 years	4	1.2
10-15 years	6	1.8
15-20 years	8	2.4
20-25 years	6	1.8
25-30 years	18	5.4
30-35 years	13	3.9
35-40 years	9	2.7
40-45 years	16	4.8
45-50 years	8	2.4
50-60 years	7	2.1
60-70 years	1	0.3
Total	333	100%

2.5 Discussion of findings derived from secondary analysis of routinely collected NHS data

Strengths and weaknesses

The aims of this secondary analysis were to map the numbers and characteristics of children and young people aged <20 years at admission, who spent one month or more in one spell in NHS hospitals in England and Scotland and see if it was possible to identify children and young people who were dependent on ventilators from routine ICD coding.

The study was successful in mapping the numbers and general characteristics, but unsuccessful in identifying children and young people who are dependent

on ventilators, from routine ICD coding. According to Williams and Wright (1998) the NHS, in common with many organisations, devotes more resources to collecting data than it does in ensuring the data it collects has some utility, or is used. Routine reports of information are known to be not as comprehensive in Britain as they are in Scandinavian countries (Williams and Wright, 1998). In the current study, it was not possible to determine the ethnicity of children and young people as 40% of the HES data was missing and ethnicity is not recorded in Scotland. This needs rectifying as there is strong evidence that children from minority ethnic communities do not receive equity of access to services (Fazil *et al.* 2002) and at present there is no robust way of monitoring access to services, or resource use.

The analysis of HES and SMR data revealed very large numbers of children and young people (over 16,000 in England and over 1,300 in Scotland) who are spending a month or more in hospital in one admission. Children's services are currently designed for children who spend on average less than 2 days in hospital (mean length of stay, 1999). There is a wealth of evidence dating back 50 years concerning the detrimental affect of prolonged hospitalisation on children and their families (for example, Robertson, 1958; Ministry of Health, 1959; Lyth, 1982; Audit Commission, 1993). The long lengths of stay reported in the current study represent considerable cost to the health service but, as stated previously, they do not indicate which children were dependent on ventilators or whether the lengths of stay were medically necessary. Nonetheless, doctors advising the study said that their own hospitals all contained ventilator-dependent children who had spent months and in some cases years waiting to be discharged home. The impact on the health-related quality of life of children will be explored further in Chapters 3 to 5, and the costs of prolonged hospitalisation will be explored further in Chapters 6 to 8.

Findings confirm that children in Scotland were not ascribed ICD codes Z99.1 or Z99.8 as a primary diagnosis or a health problem at discharge, despite the fact that colleagues in clinical settings had discharged at least 2 ventilator dependent children following prolonged periods of hospitalisation during the study period. For example, an infant with bronchopulmonary dysplasia who was

dependent on a ventilator was discharged home after an admission of over 20 months during the period 1999-2000. Analysis of the SMR data suggests that these children appear to have been classified with a primary diagnosis under ICD codes P00 - P96: Certain conditions originating in the perinatal period. If the codes Z99.1 or Z99.8 have been ascribed, it is possible that they will be found as a secondary diagnosis. A further study would be required to analyse secondary diagnostic codes.

Likewise, the English HES dataset requires the purchase and analysis of more specific data to see if ICD codes Z99.1 and Z99.8 are ascribed as either primary or secondary diagnoses at discharge. Limitations to the grant awarded meant that only the primary diagnosis at discharge was obtained. Up to 3 secondary diagnostic codes can be ascribed at discharge and this warrants further exploration. If ICD codes Z99.1 and Z99.8 are consistently ascribed as a secondary diagnosis or health problem then the size of the population could be calculated for this group of children and longitudinal studies conducted to monitor resource use over time. If diagnostic codes are not being applied appropriately then action could be taken to ensure accurate coding.

The Audit Commission report published in 2002 concurred with current findings that better quality information was vital to the NHS in order to provide complex health services to people who need them, at a time that they need them (Audit Commission, 2002c). The Audit Commission report on routinely collected data in the NHS also found that significant amounts of data were miscoded, incomplete or inaccurately coded.

The interpretation of the length of stay data for England proved particularly challenging. The specification asked for discharge data on children and young people admitted to hospital under 20 years of age. It was not envisaged that this specification would capture a significant group of very long-term patients in England who were later found to be adults who were being discharged to 'care in the community' schemes, having been admitted to hospital decades before as children. These long-term adult patients were excluded from the Scottish data.

Another major weakness of the study is that using discharge data has clear limitations. It does not give any indication of the pool of children currently in hospital awaiting discharge. Further research is needed to calculate the number of children dependent on ventilators currently in hospital.

Case definition

The varying definitions of ventilator-dependency used in the national and international literature made comparisons difficult. There is a need to standardise a simple definition of a case of a child dependent on a ventilator and to expand this definition to include valid measures of severity. In the case of respiratory failure, degree of severity could be measured by appropriate and standardised physiological and biochemical tests. International acceptance of case definition and degree of severity would allow for generalisability of incidence and prevalence across regions and countries (Williams and Wright, 1998). In addition, if a clear diagnosis of dependency on ventilation was documented in the patient notes then coders would be more likely to ascribe an appropriate code.

At present the codes for dependence on a ventilator and other enabling machines are recorded within the group of Z codes that constitute factors that 'influence health status and contact with health services'. In consideration of this growing population of ventilator-dependent adults and children, it may be appropriate to revise the ICD-10 code for dependency on a ventilator so that it appears as a primary diagnostic code further up the hierarchy. Since the ventilator is used in every instance to treat respiratory failure, one obvious conclusion would be to include dependency on a ventilator within ICD codes J00-J99 relating to diseases and health problems associated with the respiratory system.

Despite the limitations outlined, it has been possible to describe for the first time the very large numbers, general characteristics and bed use of children and young people who spend prolonged periods of time in hospital. These findings make an original contribution to the evidence base and have been used by the

Department of Health in developing a National Service Framework for children (Department of Health, 2003a).

2.6 What can qualitative research add to our understanding around how and why has this population emerged?

Review of the literature found no studies looking specifically at the reasons/factors underlying the occurrence of ventilator-dependency. Some of the narrative clinical case studies that have been written by doctors have cited advances in intensive care or the ethical framework within which doctors practiced (for example: Orłowski, 1993; Samuels, 1996).

There was some evidence in the literature that rates of ventilator-dependent children have risen concurrently with increasing investment in children's intensive care services. For example, ventilator-dependency is only a phenomenon in resource rich countries; in most middle and all low-income countries, it was reported that healthcare systems did not have resources to manage children with long-term ventilation and as a consequence therapy was discontinued and the children died (Kamm *et al.* 2001). In the UK during the 1990s, additional resources were provided to increase the number of beds for critically sick children, but no assessment of impact has been undertaken to determine if there is any relationship between the number of intensive care beds and the numbers who survive as a ventilator-dependent children.

There is a small qualitative literature exploring the factors that influence the decision-making of parents and doctors in ethically difficult situations and more specifically around ventilator-dependency.

Decision-making of parents

Qualitative studies undertaken with parents concerning the decision-making process around life support in their children have concluded that most parents clung to life as a principle good for their child; parents wanted more time for their child to experience life even when death was inevitable (Kirschbaum,

1996). A recent case brought by a mother of a disabled child with several impairments who is fighting for the right for her child to be offered long-term ventilation appears to follow a similar logic (Dyer, 2003).

Parents may have religious convictions regarding the sanctity of life and the ethics of withholding or withdrawing treatment (Kirschbaum, 1996). Pope John Paul II, however, decreed that *'man is a limited and mortal being'* and has spoken against using medical techniques to preserve a patient's life at all costs and said such medical intervention could be *'useless and not fully respectful of the patient'*. (BBC news website, 2002).

Sasaki *et al.* 2001 reported that in Japan, the healthcare system paid for the hospitalisation of ventilator-dependent children; thus they felt that the system acted as a disincentive to parents to make a decision to discontinue treatment as it was easier to continue with the status quo. As a result, Sasaki's acute neurosurgical unit had inadvertently become home to 9 severely or profoundly neurologically impaired children on ventilators, and a further 7 children and young adults on ventilators had died in recent years having lived out their lives in the neurosurgical unit.

Decision-making of doctors

In most situations in intensive care, the decision to initiate or undertake ventilation in children who go on to become ventilator-dependent is not made or planned. Even children with known neurodegenerative or respiratory disorders usually present at the hospital in a crisis, and the decision to resuscitate and ventilate has often been made in the emergency department before the child is transferred to the intensive care unit. This situation is referred to in the medical literature as the 'phenomenon of entrapment' because doctors in intensive care either have to commit to a therapy they, and possibly the family, had no control in deciding to institute, or begin the process of withdrawing ventilation with the consequence that the child will die (Orlowski, 1993). A recent study by Sritippayawan *et al.* (2003) found that in the case of children with progressive neuromuscular disease, numerous opportunities for discussing therapeutic

options with the children and their families before respiratory failure became terminal were missed or ineffective. Sritippayawan *et al.* (2003) concluded that doctors were advertently or inadvertently avoiding the subject of when it would be appropriate to discontinue treatment and institute palliative care. In contrast, other studies undertaken with doctors around ethically difficult care decisions have primarily generated concerns around the over treatment of children and the unnaturalness of invasive interventions such as ventilation (Sørli *et al.* 2000, 2001, 2003). A common theme across qualitative studies was that doctors felt that few children were seen to experience a natural death without the intervention of technology. Doctors felt that parents viewed active treatment of children as a positive act; it was easier for doctors emotionally to offer hope to parents and treat positively rather than to withdraw treatment.

Factors that shaped the exploratory qualitative fieldwork

The lack of published evidence describing the range of epidemiological reasons/factors that influenced the emergence of children dependent on ventilators constituted a significant gap that warranted further investigation. It was considered that qualitative research might add to understanding around how and why this population has emerged. Qualitative researchers assert that understanding human experience is as important as focusing on explanation, prediction and control – characteristics of quantitative research. In terms of exploring epidemiological factors around ventilator-dependency in children, qualitative research can be used as a form of social inquiry by focusing on the way doctors and nurses who care for children who are dependent on ventilators behave, interpret and make sense of their experiences and the environment within which they work. It was therefore decided to explore the reasons/factors that influenced the emergence of children dependent during a period of exploratory qualitative fieldwork.

2.7 Qualitative exploratory fieldwork

Methods

During the first 12 months of the research programme, a period of qualitative exploratory fieldwork was undertaken, including:

- Non-participant observation of activities in a children's intensive care and long-term ventilation unit in the UK (2 days a month);
- Regular face-to-face and telephone contact with health professionals and commissioners of services from all the major centres managing this group of children via the UK working party on long-term ventilation;
- Contact with relevant parents' groups, and
- Attendance at multi-disciplinary conferences concerning children and long-term ventilation.

This 12 month period of qualitative explorative fieldwork provided opportunities to talk informally with doctors, nurses, therapists, carers, children dependent on ventilators and their families. The broad aim was to explore key issues highlighted in the literature and find out the issues that were important to individuals and professional groups. Findings were used subsequently to help shape the studies reported in this thesis.

One specific question that was explored during fieldwork was how and why the population of children dependent on ventilators had emerged over the last 15-20 years? Hand written notes of informal conversations were made as soon as practical after the encounter and material such as conference abstracts were collated. Field notes and conference abstracts were subsequently analysed using thematic analysis; a rigorous process by which text is searched for related themes with similar meaning. Themes were then arranged in order of thematic significance (Holloway, 1997).

Findings

By the end of the 12 month period of exploratory fieldwork, notes from informal conversations, meetings and conferences around the emergence of the population of children had been documented. All accounts reported a consistent increase in the incidence and prevalence of children and young people dependent on ventilators over the last 10 years, although definitions around ventilator-dependency varied between individuals. For example, some doctors suggested that with morbidly obese children it was not clear if it was appropriate to define these children as dependent on ventilators when a programme of weight reduction would reverse their respiratory failure. No one had accurate evidence on the number of children and young people in the UK. Thematic analysis of interview notes revealed that this population of children emerged as an ad hoc consequence of 5 events:

1. New technology

The development of new portable ventilators that were originally designed to facilitate inter-hospital transport became available. It was never envisaged that these machines would be used outside of the intensive care unit or operated by lay people, but nonetheless these improvements in technology inadvertently opened up a possibility for home care where it had not existed before. A technological imperative, which held that because doctors can do something, doctors should do something, became established over a relatively short period of time.

2. Improvements in intensive care leading to increased survival

Improvements in intensive care meant that more children were surviving catastrophic illness and accidents but with increased morbidity, whereas in the past they would have died. This finding is supported by several accounts in the literature, such as Samuels, (1996). One example frequently cited during fieldwork was the use of the drug surfactant with very premature infants, which

has undoubtedly increased survival rates, but more babies have survived with severe lung damage and were unable to wean from ventilators.

3. Formalisation of guidance on withdrawing and withholding treatment

The formalisation of guidelines by the Royal College of Paediatrics and Child Health (RCPCH) in 1997 on withdrawing and withholding treatment in children has meant that the decision-making of doctors now occurs within a legal framework. In the past, doctors said that it was easier to make the decision not to treat a chest infection knowing that the child would most likely die quickly from respiratory failure. Currently, doctors have to establish that the child meets the criteria of one of five situations where the withholding or withdrawing of medical treatment might be considered. These are: the brain dead child; the permanent vegetative state; the no chance situation; the no purpose situation; and the unbearable situation (RCPCH, 1997). Going to Court to seek a judgement was described as an extremely stressful experience for doctors and parents. Whereas the guidance was meant to clarify the situation, in reality many doctors said they settled for a 'wait and see' approach rather than go down the judicial route. Hence it was thought by some doctors and nurses that mechanical ventilation had joined renal dialysis and artificial nutrition as technologies that could prolong life, even at a vegetative level, and in circumstances whereby the child had profound sensory and physical impairments.

Even where precedents have been established in law, doctors said that such dictums regarding withholding and withdrawal of treatment were unrealistic and out of touch with current clinical and humane practice of medicine. This was especially the case for a child who was completely alert, mentally intact but dependent on mechanical ventilation to live; the prospect of withdrawing the ventilator was said to be unpalatable to doctors, parents and children.

4. Campaigning of parents and advocacy groups

The campaigning of some parents and advocacy groups for children to be offered long-term ventilation has influenced the decision-making of doctors. The Human Rights Act (1998) has provided parents with a legal framework to assert the rights of children, especially disabled children, to equity of access to care and resources. Parents of children with life-limiting neuromuscular diseases have campaigned for their children to be offered long-term ventilation as a way of prolonging life when unable to breathe due to muscle weakness. For example, it has become common for adolescent boys with Duchenne muscular dystrophy to be supported by mask ventilation when they can no longer breathe adequately by themselves (Simmonds *et al.* 2000). Thus artificial ventilation has extended to being a palliative care intervention towards the end of life.

5. Morbid obesity: a new lifestyle disease

Doctors described big increases in the numbers of morbidly obese children who were unable to breathe effectively, especially when asleep. Put simply, fat stored around the child's neck caused obstruction of the windpipe and bulky deposits of fat limited chest wall movements and the transfer of air in and out of the lungs. Children's weight gain was attributed to increased consumption of high calorie food and reduced physical activity. These children were now treated with long-term ventilation at night or whenever asleep in order to maintain their oxygen and carbon dioxide levels within physiologically normal parameters. Respiratory failure of this nature could be reversed easily by reduction of body weight to within normal parameters for age, sex, and height.

It was also possible to map the evolutionary trajectory of usage of long-term ventilation with different groups of children and young people over the last 15-20 years.

The first children and young people were said to have emerged from neonatal and paediatric intensive care settings, including babies born prematurely with

lung disease, congenital anomalies and diseases of respiratory control that required assisted ventilation from birth or soon after birth. Of the premature babies with lung disease who went on to survive, some would eventually grow sufficient new lung tissue and come off ventilation; whereas most of the babies with congenital anomalies and diseases of respiratory control required life long ventilation, and children and young people of all ages who experienced critical illnesses, infections or accidents and who required assisted ventilation in an intensive care setting. A small number of these children never recovered their capacity to breathe unaided and went on to require life-long ventilation (such as those with serious injuries to their spinal cords and brains).

Over the last 5-8 years, with the availability of new ventilators that used a mask over the child's face to provide assisted breaths, the application of long-term ventilation has been extended to children and young people (predominantly boys) with progressive and degenerative genetic diseases such as muscular dystrophy. These children have increasingly been treated with assisted ventilation towards the end of their lives when their respiratory failure reached a critical point; the aim of assisted ventilation in this case was to prolong life. Assisted ventilation was initially started in response to a chest infection and sudden deterioration, but in the last 5 years assisted ventilation has become a prospective intervention of choice to manage progressive deterioration.

The final group of children and young people whom have emerged from what doctors described as '*nowhere*' over the last 5 years were children and young people with morbid obesity (a lifestyle disease). These children usually required mask ventilation at night and most could manage their masks and ventilators without additional need for support other than supervision from their parents.

2.8 Discussion of findings derived from qualitative fieldwork

This exploratory qualitative fieldwork presents an initial attempt to describe the reasons why this population of children has emerged and the trajectory along which new applications for long-term ventilation have become routine clinical

practice. All of the factors described are recent phenomena, including: the development of new microchip portable technology; the introduction of new drugs and therapies that have impacted on outcomes; the implementation of new guidance and the impact on doctors' decision-making; the introduction of new human and disability rights legislation and policies which advocate the involvement of parents in decision-making; and new lifestyle diseases such as obesity. Some of the findings are replicated in the literature such as improvements in intensive care medicine (Samuels, 1996; Jardine and Wallis, 1998), formalisation of the ethical framework within which doctors practice (Orlowski, 1993; Sokol, 2003), and parents' over-riding desire for their children to live (Sørliie *et al.* 2000, 2001 and 2003).

Doctors and nurses tended not to make any association between the provision of intensive care beds and resources available within the health system and the numbers of ventilator-dependent children surviving. The outcomes of increased intensive care provision warrant evaluation to establish if the unintended consequences described in the literature (that is increased numbers of ventilator-dependent children) are generalisable to the UK.

A strength of these data is that the same 5 factors were observed consistently. A weakness of this approach to data collection is that data were collected through informal exploratory fieldwork and not framed within a rigorous theoretical qualitative framework. The generalisability of findings will need to be established by the application of more rigorous theoretical approaches.

2.9 Synthesis of qualitative and quantitative findings across studies

Synthesis of findings across studies that are derived from different methodological paradigms can help to build a more complete picture of the epidemiological phenomenon under investigation (Blair *et al.* 1995). Standard epidemiological tools are important for assessing health needs of populations and qualitative findings can add an extra dimension in attempting to explain

findings in greater depth or verify quantitative epidemiological outcomes (Gough and Elbourne, 2002)

Synthesis of findings across studies confirmed that important gaps remain in the evidence available to commissioners of services. Despite concerns about methodological rigour, there was consensus across published narrative papers, three point prevalence surveys and qualitative findings derived from fieldwork, that the incidence and prevalence of children dependent on ventilators had increased markedly in the UK. None of the quantitative studies that used varying methodological approaches were able to establish with accuracy the numbers of ventilator-dependent children in the UK.

Secondary analysis of routinely collected NHS data failed to identify the same children that the doctors who participated in exploratory fieldwork work said they had discharged from their NHS Trusts during the study period. The secondary analysis established that relatively large numbers of children spent excessive amounts of time in hospital, but it was not possible to establish why they were there.

Despite methodological flaws, findings from one regional and two national surveys revealed a growth trend in the incidence and prevalence of ventilator-dependent children. Qualitative research with doctors established 5 factors as to why increasing numbers of children were surviving with ventilator-dependency. Three factors were replicated in other narrative articles (improvements in intensive care, clinical guidelines on treatment withdrawal, and parents influence on decision-making). Two factors described by doctors were new (availability of new portable technology and new lifestyle diseases such as obesity). Documenting these factors adds to the small existing literature around the epidemiology of long-term ventilation and helps explain the growth trend seen in the surveys.

Synthesised findings derived from the 4 research themes will be reviewed in an evidence based and policy context in Chapter 9 and further research directions will be outlined in Chapter 10.

2.10 Summary

Chapter 2 focused on the origins and size of the population of children dependent on ventilators in the UK (theme 1). A critique of one regional and two national surveys revealed substantial methodological flaws in both national surveys. The general trend of increasing incidence and prevalence found in the surveys was corroborated by doctors, nurses and commissioners of services who participated in exploratory fieldwork and had experienced year-on-year increases of new cases of children dependent on ventilators. The national surveys of incidence and prevalence were considered to be not sufficiently robust to be used by commissioners of services to plan future services with certainty.

The chapter went on to describe the methods and results of a secondary analysis of HES and SMR datasets to map the numbers and general characteristics of children and young people aged <20 years at admission, who spent one month or more in one spell in NHS hospitals in England and Scotland. Over 16,000 finished consultant episodes were recorded in England and over 1,300 in Scotland during 1999-2000.

It was not possible to identify children and young people who were dependent on ventilators from routine ICD coding using the primary diagnosis ascribed at discharge. Hence the study was only partially successful in meeting the aims. Synthesis of findings across studies found that there was insufficient rigorous evidence to enumerate the population of children dependent on ventilators. The growth trend in the numbers of ventilator dependent children seen in the surveys was explained in part by qualitative research with health professionals that identified five factors that had contributed to the survival of ventilator-dependent children.

The next chapter changes the focus of the thesis by moving onto the health-related quality of life of ventilator-dependent children (theme 2).

Chapter 3

Measuring the health-related quality of life of children dependent on ventilators using quantitative methods

3.0 Introduction

Chapters 3, 4 and 5 are linked sequentially and focus on reporting and synthesising findings from the empirical studies around the health-related quality of life of children dependent on ventilators (theme 2). In this chapter the health-related quality of life of children is measured using quantitative methods. Chapter 4 explores health-related quality of life in children by using qualitative methods, and in Chapter 5 a synthesis of quantitative and qualitative findings from Chapters 3 and 4 is presented.

Chapter 3 begins by discussing legal issues around the quantity and quality of a child's life; test cases are explored whereby treatment has been withheld or withdrawn from people dependent on ventilators.

The chapter then goes on to discuss the measurement of health-related quality of life as an outcome measure in health services research and reviews the few quantitative studies that have been undertaken with children dependent on ventilators. A study measuring health-related quality of life in 28 children and young people dependent on ventilators is then reported. The chapter concludes by comparing findings with published literature and describing the strengths and weaknesses of the study.

3.1 The quantity and quality of a child's life

Eiser and Morse (2001) in their review of the history and scope of quality of life measurement in children argue that there are only two outcomes that really matter to any paediatrician: the quantity of a child's life and the quality of a child's life. In a similar vein, one of the continuing debates amongst professionals that cropped up with increasing frequency during the course of the current programme of research in medical journals, conferences and exploratory fieldwork was the issue whether life on a ventilator was a life worth living. In the literature, narrative articles have been published by doctors who considered children dependent on long-term ventilation to be a failure of medical science (for example, Pilmer, 1994). A much smaller group of doctors have published narrative articles advocating on behalf of ventilator-dependent children and lobbying for services (for example, Davies, 1996). From an economic perspective there have been debates about the morality of maintaining a small population of children dependent on ventilators that consumed an exceptionally high volume of resources when services were limited by the availability of resources and other children were being denied effective treatments (For example, Hadden, 1996).

Given the polarisation of medical and public opinion as to whether life on a ventilator was worth living or not, or indeed worth paying for, somewhat inevitably, problems have arisen around decision-making when the parents or the patient themselves have not been in agreement with doctors. There have been four test cases (two in the UK and two in the United States) to deliberate on the complex ethical issues involved and another case is in the pipeline.

Test cases in law

In 1998, doctors in one fieldwork site in England went to Court in order to discontinue ventilation against the wishes of the parents of a child suffering from a severe life-limiting genetic disease. In this difficult case *re C* (a minor), the judge declared that *C* was suffering from a very severe disability and her doctor described her as being in a 'no-chance' situation. The judge deliberated that:

'...she (Child C) has such severe disease that life sustaining treatment simply delays death without significant alleviation of suffering, medical treatment may be deemed inappropriate'.

In the judgment, doctors were given leave to withdraw ventilation against the wishes of the parents and the child subsequently died with the parents still protesting an injustice had occurred (Medical Law Monitor, 1998).

More recently in the UK, the mother of a disabled 10 year old girl who suffered from epilepsy, asthma and developmental delay won the right to mount a High Court challenge to a hospital's alleged unlawful refusal to place her child on long-term ventilation (Dyer, 2003). The case was said to raise fundamental issues about the rights of disabled children to equity of access to treatment.

During an invitation only conference to discuss ethical issues and long-term ventilation in children, the author met informally with officers from the Disability Rights Commission (one of whom was a user of assisted ventilation). The view was expressed that disabled children should not be denied access to long-term ventilation on the grounds that it was not a life worth living and it was intimated that the Disability Rights Commission was likely to intervene to make submissions in the above case of the 10 year old disabled girl, with the aim of making this a test case. These complex issues were summarised subsequently in an article in the Guardian newspaper (Campbell, 2003). Campbell who is herself a user of assisted ventilation, considered that instead of helping disabled people to die, resources should be made available to enable people to live.

In complete contrast to the above position, there have been three test cases brought by users of long-term ventilation. The most recent case involved a 43 year old woman in England who was dependent on a ventilator and went to the High Court in 2002 to ask for her ventilator to be turned off as she considered her quality of life to be unacceptable (Dyer, 2002). Her doctors had refused her request, as they believed that with a programme of rehabilitation she would experience an improved quality of life although her dependency on a ventilator would remain the same. Dame Elizabeth Butler Sloss ruled that Miss B was

mentally competent to make the decision and her doctor was wrong to over-rule her wishes. Despite the ruling, Miss B had to find another doctor in an alternative hospital willing to discontinue her ventilator thereby allowing her wish to die. Warnock (2002), an independent peer and member of the Archbishop of Canterbury's advisory group on medical ethics lent her strong support to the argument that patients had no obligations to stay alive attached to ventilators just to satisfy their doctors and families.

Likewise in the United States, an alert 70 year old man who was not terminally ill but dependent on a ventilator and cared for in a hospital said that while he had no wish to die, he found it 'intolerable' being connected to a ventilator and he wished to take the risk of being disconnected (Purtilo, 1986). His doctor responded by refusing the request and subsequently applied restraints to his patient's wrists in case he should try to commit suicide by disconnecting himself from the ventilator. The doctor said that he was acting beneficently, believing that ventilator-dependency was not necessarily indicative of an unacceptably low quality of life.

The man then brought his own suit against the hospital for its refusal to honour his request to be disconnected from the ventilator. In a unanimous (but post-mortem decision, as in the meantime the patient had managed to remove the ventilator himself) three judges ruled that his refusal of treatment was 'a constitutionally guaranteed right which must not be abridged'.

Children can give consent in law when they reach the age of 16 years in England. Although a younger child can give their consent as soon as they are competent to do so, recent test cases involving children and treatment for cancer have consistently overruled the child when the position of the child is in conflict with their parents and doctors (*Gillick v West Norfolk and Wisbech Area Health Authority*, 1985; Alderson and Goodey, 1998a). Given the relative powerlessness of children in situations where decisions around treatment are contested, it is difficult to imagine a situation in the UK whereby a child would be allowed to discontinue ventilation against the wishes of their parents and doctors. There is one case in the literature whereby a 17 year old boy in the

United States expressed a wish to discontinue ventilation. He courageously dealt with all the lengthy legal issues and eventually had himself removed from the ventilator so that he could die, as he deemed his quality of life to be unacceptable (Maynard and Munth, 1987).

Can legal judgments help in defining quality of life?

Although much emphasis was given to quality of life issues in the evidence presented to the judges in these cases, it was not judgements concerning quality of life that were central in any of the deliberations. In the first case of the terminally ill baby, the judge deliberated on the legality of the Royal College of Paediatrics and Child Health guidance on withdrawing and withholding treatment in children and whether the proposed actions of the doctors were in the best interests of the child (Medical Law Monitor, 1998). The judges decided that the circumstances constituted a 'no-chance' situation and granted the doctors permission to withdraw the ventilator.

If the test case involving the 10 year old disabled girl comes to Court, lawyers intend to use the Human Rights Act (1998) to clarify whether children with disabilities (who are afforded special protection under the Act) are entitled to the same equity of access to treatments and services as non-disabled children. The case will not be brought on whether her quality of life would be improved or not by the institution of long-term ventilation.

In the test cases involving adults both in England and the United States, the cases swung on one aspect of law - whether the patients were competent to make an informed decision. In all cases they were deemed to be competent and judgement was given in their favour (that is to refuse treatment and be disconnected from the ventilator so that they could die).

More recently, the High Court in London has examined the state's obligation to protect life. In *Pretty v Department of Public Prosecutions* (2001) the Court held that while ***'the Convention does not require the state to take positive steps to force life upon the unwilling. It does require the state to take positive steps to prevent people being deprived of life'***. The Court also noted that

'the right to human dignity which is enshrined in Article 3 of the Human Rights Act is not the right to die with dignity but the right to live with as much dignity as can be afforded, until life reaches its natural end'. Again on this point of law, quality of life was not central to any of the legal arguments and the judgement raised many questions about the allocation of finite resources and whether all lives were worth living – especially as in this case the applicant, Diane Pretty, had refused assisted ventilation as she considered her quality of life to be intolerable and was asking for the right to die at a time of her choosing.

Despite the fact that the above test cases were fought on specific points of law, it is clear from the legal submissions that quality of life was an important issue to the applicants but conceptually meant different things to individual doctors, parents, campaigning bodies, adult patients and child patients. In the cases highlighted, a similar level of ventilator-dependency and impairment was considered unacceptable by some and acceptable by others. Hence, the test cases provide valuable insight into individual decision-making processes but cannot be generalised to objectively infer an acceptable quality of life for the population of people dependent on ventilators. A more systematic approach to the assessment of quality of life amongst the population of children and young people who are dependent on ventilators is needed to inform decision-making.

3.2 Ventilator-dependency, quality of life and health policy context

At the inception of the NHS, the purpose of healthcare was the cure of disease and the relief of suffering and this function was predominantly the responsibility of doctors (Foot, 1973). The World Health Organisation (WHO) definition drafted around the same time as the inception of the NHS saw health as a 'state of complete physical, mental, and social wellbeing, and not merely the absence of disease or infirmity' (WHO, 1958).

Five decades later, the situation is far more complex than predicted and the original aspirations of the NHS and simple constructs around health as defined

by WHO appear hopelessly simplistic and outdated in an era whereby in rich countries invasive medical technologies can keep a person alive with multiple impairments outside of the intensive care unit. With the introduction of new technologies, it has been shown that doctors' decision-making has been driven by legal imperatives resulting from test cases around withdrawing and withholding treatment, and equity of access to services, coupled with fear of litigation, as opposed to the outcome of empirical research evaluating the quality of life of patients.

When separated from a legal context, the aim of healthcare interventions is still to improve health and quality of life, but in the 21st century, multidisciplinary healthcare now has a broader aim to help patients to participate in the creation of new lifestyles and personal philosophies as the disease-cure model is no longer appropriate. In line with contemporary thinking, in 1980 WHO published a new conceptual model around disability called the International Classification of Impairment, Disability and Handicap (ICIDH). The new model equates to the social model of disability described previously in Chapter 1, whereby: impairment refers to any loss or abnormality of psychological, physiological or anatomical structure or function; disability is any restriction or lack (resulting from an impairment) of ability to perform an activity; and handicap is defined as the disadvantage that limits or prevents inclusion in society (WHO, 1980). The social model of disability conceptualises ventilator-dependency as a new type of lifestyle and as the test cases have demonstrated, patients who are able to do so, make their own judgements concerning what constitutes an acceptable quality of life (Noreau and Shepherd, 1995). From a methodological perspective, it was decided that quality of life should be used as an important dimension in determining the benefit of long-term ventilation as an intervention.

Health-related quality of life as an outcome measure

During exploratory fieldwork the need to evaluate health-related quality of life as an outcome was highlighted as a priority by doctors, children and parents alike. Doctors prioritised the need to undertake some quantitative measurement whereas children and parents urged that their views and experiences should be

explored via consultation with them. It was therefore decided to include both qualitative and quantitative methods to evaluate health-related quality of life and undertake some methodological work to synthesise the findings.

The remainder of this chapter will concentrate on quantitative methods of exploring health-related quality of life and describes the methods and findings concerning a quantitative study with 17 ventilator-dependent children and 28 parents.

3.3 Quantitative measurement of health-related quality of life

Purpose of health-related quality of life measures

The general purpose of health-related quality of life measures is to discriminate levels of functioning between groups of children and to detect changes in function over time. Results derived from psychometric measures are designed to arrange children along a continuum of function or wellbeing (Eiser and Morse, 2001). From a health services research perspective, the measurement of health and quality of life is central to the evaluation of healthcare. By observing the extent of changes in health and the impact of health on quality of life, the benefits and disbenefits of healthcare for both individual patients and groups of patients can be evaluated (Kind *et al.* 1998).

Guyatt *et al* (1993) proposed that health-related quality of life measures can be used for three purposes: to discriminate the quality of life between individuals and groups (discriminative index); to predict or classify individuals into a set of predefined categories (predictive index); or to evaluate or assess the magnitude of longitudinal change in an individual or group (evaluative index).

In relation to ventilator-dependent children, quality of life measures may thus be used to determine how far the quality of life of ventilator-dependent children differs from other children. Second, it may be important to be able to classify ventilator-dependent children in order to provide appropriate educational and social opportunities, and third, measures can be used to evaluate the impact of different treatments or interventions over time. It may however be unreasonable

to expect that an intervention would lead to changes in functional capacity for all children, but more realistically an intervention might lead to broader social benefits, such as increased confidence, or ability to enjoy a wider range of activities.

Model of disability

Theoretically, measurement of health-related quality of life is underpinned by the medical model of disability. This is particularly the case for generic health status and quality of life tools whereby the tools were developed and tested with populations of 'normal' children. Validation studies provide normative reference values so that the findings of subsequent studies can be compared with 'normal' populations of children of a similar age and gender.

Methodological issues in the measurement of health-related quality of life in children

Over the past 20 years several generic and disease specific measures have been developed to measure health and health-related quality of life in children (Eiser and Morse, 2001). Measuring health-related quality of life in children presents a number of methodological challenges. Adult proxies (usually parents and healthcare workers) are frequently used in data collection. When proxies provide data about health conditions responses can be strongly influenced or distorted by how much of a burden the child and their care are to the proxies as well as how important various outcomes are to them. Assessments by physicians and other healthcare workers have been shown to be highly inaccurate when compared to the child's own perception (Eiser and Morse, 2001).

A second issue concerns who determines what domains of childhood define quality of life (Sennhauser, 1997). Conceptually, it is the individual child's perspectives that are the most important, but a disease-specific or group-specific measure has not been developed with children dependent on ventilators and therefore it is not known what is important from the perspective

of the children themselves. It may be that for these children, many of who have profound physical impairments, that the mechanics of task performance may be less important than the ability to enjoy life.

The third challenge concerns the developmental changes during childhood, which makes it impossible to apply any single measure to all age groups from neonates to adolescents. In addition, many ventilator-dependent children have intellectual and physical impairments that may affect their ability to understand and complete the questionnaires.

What is already known about health-related quality of life of children dependent on ventilators derived from quantitative measures?

Although an increasing number of ventilator-dependent children are surviving and eventually being cared for at home, few studies in the literature measure their health-related quality of life from multiple informant perspectives. Most studies have focused on ventilator-dependent adults and from a methodological point of view it is unclear if these data can be extrapolated to children. Hence studies undertaken solely with adults are not included here.

Studies that compare the perceptions of ventilator-dependent children with parents and healthcare workers

Two studies compared children's self reported health-related quality of life with that of their parents (Paditz *et al.* 2000 and Lumeng *et al.* 2001).

Paditz *et al.* (2000) developed a questionnaire for children and their parents to determine if they had different perspectives on health-related quality of life and to what extent children and their parents accepted home ventilation as a means of improving their quality of life.

The questionnaire was administered to 62 German children with a mean age of 14.8 years and their parents. Sixty children were ventilated via nasal mask and 2 via a tracheostomy. The findings revealed that evaluation of 9 out of 24 items was significantly different between parents and children. Compared to their

parents' perception, children contemplated less about their ventilator, felt more satisfied with their life, thought less frequently about death and dying, had fewer headache episodes, said that the nasal mask was less uncomfortable, felt less unhappy, felt less bored, and felt better than their parents thought.

Although the findings are interesting and reinforce that children have different perceptions from those of their parents, this study has a number of obvious methodological and statistical weaknesses. The team developed their own health-related quality of life instrument. The instrument is not named and psychometric properties are not given. The sample of children almost all used mask ventilation at night and were not dependent on ventilation 24 hours a day. This sample does not therefore represent the range of ventilator-dependency and impairments demonstrated by the population as a whole and described in Chapter 1. Correlation coefficients were not given so it is not possible to determine the degree of association between parent's and children's scores. Finally, the analysis did not differentiate between children and adolescents. This is important as adolescents may differ in important ways from both younger children and adults.

Lumeng *et al.* (2001) formulated their own questionnaire to look at the capability for independent living and an assessment of wellbeing in ventilator-dependent children. Questionnaires were received from 38 children and or their parents over a 12 year period in the out-patient department of their hospital in the United States. The children ranged from 7 months to 19 years and represented the range of ventilation modalities and levels of disability. Questionnaires were only administered to children over 12 years of age.

The study reported that the majority of children were satisfied or very satisfied with how they spent their time. Children over 12 years old were reported to be significantly less satisfied with their daily activities than children under 12 years. Older children were found to be significantly less emotionally content, which Lumeng *et al.* put down to differences in reaction to ventilator-dependency. Unlike other studies undertaken with children, there were no significant differences between the responses of parents and children. Correlation

coefficients are not given so it is not possible to determine the degree of association between parent's and their children's scores

Unfortunately this study also has several methodological flaws. Primarily, the researchers developed their own tool and did not test reliability or validity. Children under 12 years were not surveyed and proxy estimates from their parents are compared with older children's estimates. No differences were found on any dimension between parents and children, which may indicate that the instrument was not sensitive enough to pick up subtle but important differences. Finally, the findings were not compared with reference values from an age appropriate population of school children so conclusions cannot be drawn concerning age-related changes in emotional state.

Neither of these two studies used instruments that met the most frequently cited requirements of an acceptable measure of quality of life (reliability and validity) and as such findings cannot be considered reliable or generalisable (Eiser and Morse, 2001).

Health and quality of life of children dependent on ventilators derived from studies with doctors and/or parents

The study by Baldwin-Myers and Oppenheimer, (1996) was identified prior to, Sasaki *et al.* (2001) was published during and Bach *et al.* (2003) was published following completion of the empirical work reported here.

Baldwin-Myers and Oppenheimer (1996) measured quality of life and quality of care at 2 points (pre and post discharge from hospital) with a group of 91 long-term hospitalised ventilator-dependent patients in California who were subsequently discharged home. The sample included adults and parents of children but no demographic information is given and findings do not differentiate children from adults. The researchers designed their own tools to measure quality of life and quality of care, and no data is available regarding their reliability or validity.

Sasaki *et al.*, (2001) measured quality of life in 45 children (aged 1-21 years) with neuromuscular disorders who underwent long-term life sustaining ventilation in their hospital ward in Japan during the period 1990-2000. Quality of life was measured by doctors using 2 scales designed by the researchers. Daily life status was measured on a scale of 1 to 5 (ranging from 1 – patient stayed at home to 5 – almost always bedridden) and communication ability was measured on a similar scale (ranging from 1 – normal speech to 5 - no communication). Five patients were reported as being able to spend some time during the day at home. The remaining children spent almost all day in bed and most had severe sensory impairments and little communication.

Bach *et al.* (2003) compared 46 United States healthcare professionals' assessments of the quality of life of children with a progressive degenerative condition, who used non-invasive ventilation via mask or tracheostomy, with 46 parents. Bach *et al.* developed their own tool using Likert scales to assess 6 quality of life issues (child's quality of life, your quality of life, effort to care for child, how happy is the child? and is the child's life worth living?), and 10 semantic differentials.

The study found that healthcare providers had significantly lower estimates of the children's quality of life than the children's parents. Correlation coefficients are not given, so it is not possible to determine the degree of association between parents' and professionals' scores. Parents and care providers found life with the children to be satisfying, although the effort parents felt in raising the children was high. Primary caregivers in particular experienced a high level of burden. Only 45% of clinicians said that they would recommend ventilation as an intervention to prolong the lives of these severely impaired children.

This study has serious methodological flaws; Bach *et al.* developed their own tool and did not provide data on reliability or validity, and few demographic data were given concerning the sample. Although the findings are difficult to generalise, the study does add weight to findings derived from other patient groups, that healthcare professionals rate quality of life as lower than parents or the children themselves (Rothwell *et al.* 1997).

None of the instruments used by Baldwin-Myers and Oppenheimer (1996), Sasaki *et al.* (2001) and Bach *et al.* (2003) fulfilled the criteria of an acceptable measure of quality of life (Eiser and Morse, 2001).

Quality of life of families

The studies with the largest samples are those that aim to evaluate the quality of life of ventilator-dependent children and have done so by assessing family and caregiver stress as opposed to the health and quality of life of the child *per se*.

Aday and Wegener (1988) conducted a mixed method telephone survey with parents of 141 ventilator-dependent children aged 1-21 years in the United States. At the end of a semi-structured interview, 4 validated instruments (Functional status, PARS II, Impact on Family and Caregiver Wellbeing) were administered to parents. Unfortunately, this study is not well reported and the quantitative findings are not made explicit amongst the qualitative findings, making comparisons with other studies using similar instruments impossible.

Quint *et al.* (1990) measured the impact on families of caring for a child dependent on a ventilator in California. A validated measure (Impact on Family Scale) was administered to parents of 18 children aged 1-21 years. Unlike Bach *et al.* (2003), analysis of scores showed no differences in impact between primary care givers and their spouses. Primary caregivers, however, showed significantly reduced coping with a longer duration of home care. Correlation coefficients are not given so it is not possible to determine the degree of association between primary caregivers and their spouse's scores. This study is limited by the small sample, and findings are not compared with normative values derived from a reference population.

Other studies conducted in the United States attempt to measure the impact of caring for a range of technology-dependent children at home within the same sample (For example, Patterson *et al.* 1992; Leonard *et al.* 1993; Ray and Ritchie, 1993; Teague *et al.* 1993; and Fleming *et al.* 1994). These studies use a range of measures and do not differentiate between children dependent on

ventilators and those dependent on less complex technologies, which makes comparison with other studies concerning only children dependent on ventilators difficult. Hence, no further evaluation of these studies is presented here.

Factors that influenced the quantitative empirical study

During exploratory fieldwork with doctors who care for ventilator-dependent children and their families, it became obvious that almost all doctors valued quantitative approaches to measuring health-related quality of life more highly than qualitative approaches. They were familiar with interpreting outcomes derived from structured instruments and were keen for a quantitative study of health-related quality of life to be undertaken.

Review of the literature revealed that little research to measure the health-related quality of life had been undertaken with children dependent on ventilators and the few studies that had been undertaken were beset by conceptual problems and had serious methodological flaws. None of the studies were conducted in the UK and only 2 small studies (Paditz *et al.* 2000; Lumeng *et al.* 2001) attempted to compare the children's own perceptions of their health-related quality of life with those of their parents and/or caregivers. The low volume and low quality of the literature meant that it was not possible to perform a meta analysis of outcomes. This lack of rigorous evidence around the measurement of health and health-related quality of life of children dependent on ventilators represented a significant gap in the evidence base and warranted further investigation. It was therefore decided that it would be a priority to undertake some research with children and their parents and offer some recommendations for improving future research activities designed to measure health-related quality of life in this group of children.

Members of the advisory group set up to guide this part of the empirical work recommended that it would be advantageous to survey the entire population of ventilator-dependent children entered onto the national database (Jardine *et al.* 1999) as described in Chapter 1. However, this approach presented a number

of ethical and technical dilemmas. Primarily, it was not clear if parents who consented to have their children registered on the database had also consented to their data being used for research purposes. The original consent form that parents signed contained no reference to the use of their data for research purposes.

Even if the ethical issues around access could be overcome, there was uncertainty about the impact of parents helping their children to fill out the questionnaire. During exploratory fieldwork, it became obvious that a number of children would need help to complete the questionnaire and there was a worry that parents may knowingly or unknowingly influence their children's responses. Finally, the database had been assembled as a point prevalence survey and there was no way of knowing if any of the children had died subsequently. It would have been inappropriate and insensitive for parents to receive a questionnaire concerning their child's health-related quality of life after their death.

Given these logistic and ethical issues, it was decided not to undertake a survey of health-related quality of life with the entire known population of children, although it was acknowledged as being likely to generate data from greater numbers of children than the approach subsequently adopted. As a compromise, it was decided to include the measurement of health-related quality of life as part of the case studies with 35 index children and their families. Although this approach would potentially yield fewer completed questionnaires, the data would be rigorously collected in an ethical manner by a single researcher who could help the children and young people to communicate and record their responses as impartially as possible. As it would only be feasible to measure health-related quality of life on one occasion, it was decided to use measures of health-related quality of life as a discriminatory index to determine if there were differences between ventilator-dependent children, their parents, and normative populations of school children.

3.4 Quantitative study to measure health-related quality of life in ventilator-dependent children

Methods

Aim

The aim was to measure the health and health-related quality of life of a sample of ventilator-dependent children age 4-19 years and compare the findings with those of their parents and normative values derived from a representative sample of children in the general population.

It was decided to focus the study on three areas of interest which are reflected in the following research questions:

1. Do ventilator-dependent children have lower health-related quality of life than a normative population of school children?
2. Do ventilator-dependent children report lower health-related quality of life than the proxy estimates reported by parents?
3. Do ventilator-dependent children living in hospital report lower health-related quality of life than ventilator-dependent children living at home?

Choice of tools

Review of the literature highlighted that few studies had used validated instruments and as a result were methodologically flawed. During the planning stages, Eiser and Morse published a Health Technology Assessment systematic review of quality of life measures in chronic diseases of childhood (Eiser and Morse, 2001). This comprehensive and rigorous systematic review advised against developing a specific tool for a small population where it would be very difficult to test the psychometric properties of the measure. Given the fact that children dependent on ventilators constituted a small population, it was decided that there were insufficient children with which to develop and test a group-specific instrument.

Consideration was also given to the Patient Generated Index which was originally developed for adult patients with chronic lung disease (Ruta *et al.* 1994). The Patient Generated Index aims to quantify the effect of a medical condition on a patient's quality of life in a way that has meaning and relevance in the context of their daily lives. The instrument is completed in three stages. First, patients are asked to identify the five most important areas of their lives affected by breathing problems. Second, they are asked to rate how badly they are affected in each priority area identified on a scale of 1 to 100, where 0 represents the worst they can imagine and 100 represents exactly as they would like to be. Finally, patients are asked to imagine that they can improve some or all of their priority areas and are given 'points' to spend across one or more areas that they would like to improve.

Although the underlying philosophical approach of the Patient Generated Index was appealing, it was decided not to use this instrument as it had not been used with children and normative data were not available. More specifically, some ventilator-dependent children are known to have experienced lives characterised by social exclusion (Noyes, 1999). It was not known what impact social exclusion would have on the ability of a child to complete the Patient Generated Index. Macduff and Russell (1998) have reported considerable difficulties in using the Patient Generated Index with a population of disabled adults in a study whereby only 12% of subjects were able to complete the three stages of the instrument. Likewise, some ventilator-dependent children have sensory and communication impairments ranging from mild to severe which would limit their ability to complete the required tasks. Finally, the tool would be too complex with too high a Flesch score for children under 14 years of age with age appropriate cognition (Flesch, 1918).

In identifying appropriate children's generic instruments, reference was also made to a doctoral thesis containing a systematic review of measures of health and health-related quality of life for use with children following discharge from paediatric intensive care units (Granger, 2001). The author had acted as a second reviewer for this systematic review and had evaluated all disease-specific and generic measures of health-related quality of life in children.

Granger's systematic review (2001) revealed that there were few generic measures of health and quality of life designed for use with young children and babies. The theoretical sample planned for the case studies reported here included a group of children under 5 years old. Given the wide age range of children in the sampling frame (from infancy up to 19 years) it was decided that a compromise was required, as no appropriate measure covered the entire age range. It was considered more important to concentrate on measuring health-related quality of life in children from age 4-19 years (as opposed to children age 0-3 years), as a validated range of measures were available for use with this group.

Based on Eiser and Morse's (2001) and Granger's (2001) systematic reviews and advice received from the advisory group, a modular questionnaire was formulated from 2 complementary tools: the EQ-5D paediatric version (EuroQol Group, 1990) and the KINDL (Bullinger *et al.* 1994). The 2 tools measured different aspects of health-related quality of life: the EQ-5D being a generic measure of health status and the KINDL a generic measure of health-related quality of life.

EQ-5D paediatric version

The EQ-5D was originally developed for use in evaluating health and healthcare of adults. A paediatric version has been developed and the reliability and validity has been tested with a group of over 3000 school children aged 7 to 19 years. It is cognitively simple and takes only a few minutes to complete. The EQ-5D describes health status in terms of 5 dimensions, including: mobility; self-care; usual activity; pain/discomfort; anxiety/depression. Each dimension is divided into 3 levels: no-problem, some problem, and extreme problem. In addition, children are asked to give a rating of their overall health status using a thermometer-like scale marked 0-100. By combining different levels from each dimension, the EQ-5D defines a total of 243 health states. It produces a numeric score for health status on which full health has a value of 1 and death a value of -0.59.

On the advice of the EuroQol group, it was decided to make a small adaptation to the EQ-5D. In addition, respondents were asked to give a rating of their (or their child's) overall quality of life using a thermometer-like scale marked 0-100, to see if they made a distinction between overall health and quality of life. They were also asked whether their (or their child's) health and quality of life was better, worse, the same, or much worse than other children of the same age.

The original paediatric EQ-5D was only administered to children. For the purposes of this study it was decided to include a parent version to see if responses varied between parents and their ventilator-dependent children. A copy of the EQ-5D can be found in Appendix 1.

KINDL

To complement the EQ-5D that measures health status, the KINDL was chosen, as it is a generic measure that assesses 4 domains of quality of life in children aged 4-16 years and where appropriate their parents. The tool measures psychological wellbeing; social relationships; physical functioning; and everyday life activities (thereby not just relying on functional status). The KINDL contains an additional sub-scale called 'disease' which can be completed by children who have a chronic illness or have experienced prolonged hospitalisation. This additional sub-scale aims to measure the child's quality of life with respect to their illness. Age-appropriate versions of the KINDL are available for children aged 4-7 years, 8-12 years and 13-16 years. The KINDL has been shown to have a high degree of reliability and validity (Ravens-Sieberer and Bullinger, 1998).

The KINDL has also been produced in a computer format, which appealed as a method of administration for the children and young people who do not use speech to communicate. During exploratory fieldwork, it was observed that the children were familiar with using technology of this nature in their everyday lives and for many children with certain types of spinal and head injuries, it was the only way that they communicated. However, when the computer programme arrived it was discovered that there were several grammatical mistakes in the German to English translation. This was an unexpected problem as the KINDL

had been used widely in English studies. For example, one item stated 'I avoided others to notice my illness' and other items talked about attending kindergarten. The grammatical and cultural errors could be easily corrected in the paper format, but the computer programme could not. The computer format was tried out with a child, but they disliked the fact that the researcher was hovering over their shoulder ready to point out errors in the English translation. The child also appeared phased by the grammatical errors when they were pointed out and the errors themselves became the focus of their attention. Although the software itself was evaluated highly, it was decided not to use it in the empirical study because there was insufficient time for the KINDL team to correct the content.

The KINDL questionnaire consists of 24 Likert-scaled items. Children aged 4-7 choose from 3 response categories (never, sometimes and very often), whereas older children choose from 5 response categories ranging from never to always. The KINDL has been used and tested with over 3000 healthy and chronically ill children and groups of parents. The psychometric results revealed a high degree of reliability (Cronbach's $\alpha \geq 0.70$ for most of the sub-scales and samples) (Ravens-Sieberer and Bullinger, 1998). In terms of convergent validity, the KINDL sub-scales have been correlated with both the subjective health dimensions of the Child Health Questionnaire (Landgraf *et al.* 1998), and the SF-36 (Bullinger and Kirchberger, 2000). It has been shown that the KINDL can discriminate between differences in the impairment of health-related quality of life in different diseases (asthma, atopic dermatitis and obesity) both on a sub-scale level and in terms of the total score (Ravens-Sieberer and Bullinger, 1998). Copies of the age appropriate questionnaires for children together with the parents' versions can be found in Appendix 1.

Ethical issues

Permission was obtained from the German KINDL and EQ-5D authors to use their instruments. Ethical approval was obtained from one MREC and 9 LRECS to interview and administer the KINDL and EQ-5D to children and their parent(s).

An advisory group including a young ventilator-dependent person, parents, relevant health, social and education professionals, and researchers was convened to support the study. Time for debriefing with a qualified social worker following data collection was built into the schedule to deal with any aspects of disclosure or child protection (Thompson 1990, Steward *et al.* 1993). All young people were asked for their consent to participate, assured of confidentiality and if unable to give consent in law, parents or legal guardians were asked to sign an additional consent form. All children and their families were presented with a copy of the findings and a report was written for younger children tailored to their individual needs.

Training

The author underwent additional training to develop further skills in interviewing children who used non-verbal methods of communication (Puddicome, 1995; NSPCC, 1997). As part of the MRC Fellowship, additional training was received from the Cochrane Health-Related Quality of Life Methods Group in the design and conduct of a study to measure health-related quality of life.

Design and sample

The EQ-5D and KINDL were included as part of a case study with 35 index children who were dependent on ventilators.

It was planned to undertake case studies concerning 35 index children and young people who were ventilator-dependent and aged from 1 year to 19 years. The children and their families were recruited via 11 hospital consultants in the UK. To ensure that the cases represented a range of children and their families, it was decided to develop a theoretical sampling strategy in order to minimally include the following demographic characteristics amongst the index cases and those interviewed:

- 10 children age 5 and under;
- 10 children age 6-12 years;
- 10 children and 13-19 years;

- Representation from both boys and girls in each age band;
- Representation from children who required assisted ventilation for 24 hours per day and children who used it for less than 24 hours per day in each age band;
- A mix of children who lived in hospitals, domestic settings and 'looked after' children in the care of social services in each age band, and
- Representation of parent(s)' views.

Recruitment was successful, apart from achieving representation of 'looked after' children in each age band. The reasons for this are explored further in Chapter 3.

Questionnaires were administered where appropriate to children age 4 -19 and parents of children age 4-19 years. The resulting subset, which excluded children under 4 years of age, is shown in Figure 3.1. This subset included 17 ventilator-dependent children, and parents of 28 ventilator-dependent children (11 girls and 17 boys); including 17 matched pairs of ventilator-dependent children and their parents. Four children lived primarily in hospital and the remainder lived in domestic settings. Ten were dependent on ventilation 24 hours per day and 18 less than 24 hours.

Figure 3.1. Demographic information concerning the sample.

Age range	No	Sex	Diagnosis	Type of ventilation	Location of child	Lone parent family	Ethnic minority	Looked after child	Child Questionnaire	Parent Questionnaire
4-12 Yrs	1	F	Spinal injury	PPV trachesotomy (24 hours)	hospital	✓	✓			✓
	2	M	Congenital	PPV (when asleep)	home	✓	✓			✓
	3	F	Congenital	PPV mask (night time)	home					✓
	4	F	Congenital	PPV tracheostomy (when asleep)	home		✓			✓
	5	M	Spinal/head injury	PPV trachesotomy/PPD (24 hours)	hospital					✓
	6	M	Congenital	PPV mask (Night time)	home	✓			✓	✓
	7	M	Congenital	PPV tracheostomy (24hours)	home					✓
	8	M	Congenital	PPV mask (when asleep)	home					✓
	9	F	Spinal/brain injury	PPV mask (when asleep)	home				✓	✓
	10	M	Congenital	PPV tracheostomy (when asleep)	home	✓				✓
	11	F	Congenital	PPV mask (when asleep)	home				✓	✓
	12	F	Spinal/brain injury	PPV tracheostomy (24 hours)	hospital				✓	✓
	13	M	Congenital	PPV mask (when asleep)	home				✓	✓
	14	M	Spinal/brain injury	PPV tracheostomy/PPD (24 hours)	home				✓	✓
	15	M	Congenital	PPV mask (night time)	home	✓				✓
	16	M	Congenital	PPV trachesotomy (24 hours)	home		✓		✓	✓
	17	M	Congenital	PPV tracheostomy (24 hours)	home		✓		✓	✓
13-19 yrs	18	M	Congenital	PPV mask (night time)	home				✓	✓
	19	M	Spinal/brain injury	PPV tracheostomy (24 hours)	home		✓		✓	✓
	20	F	Congenital	PPV mask (night time)	home				✓	✓
	21	F	Congenital	PPV mask (when asleep)	home	✓				✓
	22	F	Congenital	PPV mask (night time)	home					✓
	23	M	Congenital	PPV nasal pillows (night time)	home	✓			✓	✓
	24	M	Congenital	PPV mask (night time)	home				✓	✓
	25	F	Congenital	PPV mask (night time)	home				✓	✓
	26	M	Congenital	PPV mask (night time)	home				✓	✓
	27	F	Spinal/brain injury	PPV mask (night time)	home				✓	✓
	28	M	Spinal/head injury	PPV trachesotomy (24 hours)	hospital				✓	✓

Key: PPV: positive pressure ventilation PPD: Phrenic pacing device

Administration of the questionnaire

The questionnaire was administered at a convenient time during an interview with the child and or their parents where the child was currently living. Parents filled out the questionnaire by themselves. Children and young people who were able to, filled out the questionnaire by themselves. The author offered support to children who needed help with reading the questions and filling in responses. In situations where parents and children were both asked to complete questionnaires, they were asked not to confer.

Data analysis

Data were entered into an Access™ database (Microsoft, 2000) and then transferred into SPSS™ version 11 (SPSS, 2001) for the purposes of analysis. Data analysis packs were provided for both measures, which included a computerised analysis programme and normative reference data derived from large studies with school children.

The KINDL analysis programme automatically replaced missing values by an estimate, provided the individual had answered at least 70% of the items on the sub-scale. In practice, however, there was minimal missing data on the returned questionnaires.

The EQ-5D questionnaire was analysed by converting the self-reported description derived from each of the 5 dimensions, into a health state. A total of 243 health states are defined in this way. Each health state is referred to in terms of a 5-digit code, which is converted to a score (ranging from 1 indicating perfect health to -0.59 worst possible health) using tables of values provided (EuroQol, 1998). The thermometer generates a self-rating of current health-related quality of life with 100 being best imaginable health state and 0 the worst imaginable health state. The score is used with the 5-digit health state classification to build a composite picture of the child's health status. Values for the 243 health states are based on data collected from a sample of 3,000 school children in the UK.

The KINDL questionnaire was analysed by adding the item responses marked on each sub-scale. The sub-scale scores are then transformed to values between 0 and 100 with higher scores representing better health-related quality of life (Ravens-Sieberer and Bullinger, 2000).

Normative reference data for the KINDL and EQ-5D indicated that responses from children varied with age. The KINDL analysed data for all children and also presented analyses for 2 subgroups (children age 4-12 and 13-19). For consistency, the analysis described here was undertaken in a similar way.

Analysis of the KINDL and EQ-5D compared the differences between all ventilator-dependent children (4-19 years) and 2 subgroups of ventilator-dependent children (4-12 and 13-19 years) with parents, populations of school children, and in the case of the KINDL, populations of children with other chronic illnesses. In addition children living in hospital were compared with children living at home. It was hypothesised that ventilator-dependent children would report more health problems and a lower quality of life than a population of school children, parents would report more health problems and a lower quality of life than their ventilator-dependent children, and children living in hospital would report a lower quality of life than children living at home.

Choice of statistical tests and potential limitations to the interpretation of findings

The statistical tests were chosen with the specific research questions in mind.

Question 1. Do ventilator dependent children have lower health-related quality of life than a normative population of school children?

Question 2. Do ventilator dependent children report lower health-related quality of life than the proxy estimates reported by their parents?

Question 3. Do ventilator-dependent children living in hospital report lower-health related quality of life than ventilator-dependent children living at home?

Comparison of groups

One sample and paired sample t tests were used to test for group and subgroup differences.

Estimation of association

It was decided to assess whether children's and parent's scores were associated. As the two comparator scores were continuous variables, Pearson's correlation coefficient was used to measure how children's and parent's scores were related and the relative differences between scores. However, Pearson's correlation coefficient only measures the degree of association between the two scores and does not measure how closely they agree (Altman, 1991).

Prediction

It was also considered important to compare the observed and expected frequency of the individual domains that make up the health-related quality of life questionnaire (for example, observed versus expected frequency of mobility problems amongst ventilator-dependent children compared with a normative population of schoolchildren). The Chi-Square statistic was used for this purpose (Altman, 1991). It was however, anticipated that as ventilator-dependent children are known to have severe problems with mobility in comparison to a general population of school children that the P values would show very significant differences between observed and expected frequencies.

Limitations concerning the interpretation of data

The use of the t test is based on the assumption that the data for each group (with independent samples) or the differences (with paired samples) have an approximately normal distribution. Similarly, the correlation coefficient can be calculated for any dataset, however there is a restriction on the validity of the associated hypothesis test, which is that the two variables are observed on a random sample of individuals and that the data for at least one of the variables have a normal distribution in the population. For the calculation of a valid confidence interval for r both variables should have a normal distribution. In practice, therefore, it is preferable for both variables to have approximately normal distribution for any use of Pearson's r . Data of this type will display a roughly elliptical pattern, with a degree of elongation of the ellipse being related

to the correlation coefficient. For small samples such as the one analysed here, normal distribution may be hard to detect, so where appropriate a scatter diagram of the data was produced to show whether the data show a reasonably elliptical pattern.

Correlation analysis is also especially sensitive to sample selection because the between subject variation in each variable enters directly into the calculation (Altman, 1991). As indicated above, there is an implicit assumption that the ventilator-dependent children in this study constitute a random sample (or nearly so) from a specified population of individuals (in this case the entire population of ventilator-dependent children in the UK). It may therefore be potentially misleading to calculate the correlation when the sample comprises different subgroups (such as morbidly obese children and children with spinal and/or brain injuries who both need assisted ventilation but have varying degrees of morbidity). Another consequence of the mixing of subgroups is that the data when mixed may not be normally distributed, but the effect cannot be detected unless the groups are very different and the sample is large.

Although the purposive sampling strategy described previously was designed to reflect the known population of ventilator-dependent children on the UK database, there is a danger that including or excluding individuals from the sample may have an important effect on the correlation. Therefore, with such a small sample, restrictive sampling may be a cause for concern and results should be interpreted with caution until data derived from a larger sample of children is available for analysis.

Results

The findings from each instrument will be presented separately.

KINDL: Children aged 5-12 years

Data were obtained from 8 ventilator-dependent children and 17 parents, and scores were compared with normative reference values from 918 school children of a similar age. Findings are summarised in Table 3.1

Ventilator-dependent children compared with school children aged 5-12

As predicted, Total Quality of Life scores for ventilator-dependent children age 5-12 years were significantly lower than for school children ($P=0.02$).

Of the 7 domains that contribute to the total score, only Physical wellbeing of ventilator-dependent children was significantly lower than school children ($P=0.004$), although 5 domains nearly reached significance.

Table 3.1. Summary of KINDL results: Children aged 5-12 years. Original in colour.

	Normative Reference value School Children Aged 4-12 N=918						Ventilator-dependent children Aged 5-12 N=8						Parents of ventilator-dependent children Aged 5-12 N=17						Mean differences: Ventilator-dependent children and School children	Mean differences 17 parents and 8 ventilator- dependent children	Mean differences 8 parents matched with ventilator- dependent children																		
	Boys			Girls			Total			Boys (n=5)			Girls (n=3)			Total						Boys (n=11)			Girls (n=6)			Total											
	Mean	SD		Mean	SD		Mean	SD		Mean	SD		Mean	SD		Mean	SD					Mean	SD		Mean	SD		Mean	SD		Mean	SD							
Total Quality of Life Score	76.67	8.66		76.83	8.63		76.75	8.63		70.20	7.08		59.65	14.44		66.25	14.44		62.44	9.03		69.43	14.87		65.06	14.87		62.44	9.03		69.43	14.87		65.06	14.87		Ventilator-dependent children had significantly lower scores than school children P=0.02 (-19.58 to -1.41)	NS	NS
Physical wellbeing	76.68	13.03		74.43	14.19		75.55	14.19		56.25	13.97		56.25	0.00		56.25	0.00		49.37	19.86		77.08	8.53		59.76	8.53		49.37	19.86		77.08	8.53		59.76	8.53		Ventilator-dependent children had significantly lower scores than school children P=0.004 (-29.85 to -8.74)	NS	NS
Emotional wellbeing	82.89	10.67		83.11	11.33		83.00	11.33		73.75	16.17		87.50	0.00		77.67	0.00		64.37	15.03		69.79	22.50		66.40	22.50		64.37	15.03		69.79	22.50		66.40	22.50		Ventilator-dependent children had significantly lower scores than school children P=0.004 (-29.85 to -8.74)	NS	NS
Self-esteem	66.52	18.95		66.68	17.83		66.60	17.83		62.50	16.29		50.00	17.67		61.60	17.67		58.12	18.41		68.75	29.58		62.87	29.58		58.12	18.41		68.75	29.58		62.87	29.58		Parents had significantly lower scores than children P=0.04 (-18.57 to -0.39) Correlation r=0.78, P=0.03	NS	NS
Family	83.58	13.14		84.40	12.85		83.99	12.85		76.25	6.84		68.75	44.19		74.10	44.19		68.12	13.32		80.20	10.01		72.65	10.01		68.12	13.32		80.20	10.01		72.65	10.01		Parents had significantly lower scores than children P=0.04 (-18.57 to -0.39) Correlation r=0.78, P=0.03	NS	NS
Friends	78.21	12.78		78.10	13.78		78.15	13.78		85.00	7.12		53.12	39.77		75.89	39.77		66.25	10.70		66.66	25.81		66.40	25.81		66.25	10.70		66.66	25.81		66.40	25.81		Parents had significantly lower scores than children P=0.01 (-27.85 to -4.42)	NS	NS
School	72.35	12.88		74.10	12.29		73.21	12.29		63.75	12.80		68.75	0.00		64.58	0.00		65.62	28.70		65.62	28.70		66.51	28.70		65.62	28.70		65.62	28.70		66.51	28.70		Parents had significantly lower scores than children: P=0.01 (-27.85 to -4.42)	NS	NS
Disease*							78.67			87.50	13.81		62.50	39.74		78.12	39.74		57.50	21.31		69.44	22.92		61.97	22.92		57.50	21.31		69.44	22.92		61.97	22.92		Parents had significantly lower scores than children: P=0.01 (-27.85 to -4.42)	NS	NS

* Derived from a population of 1050 chronically ill children

Gender differences: Children aged 5-12 years

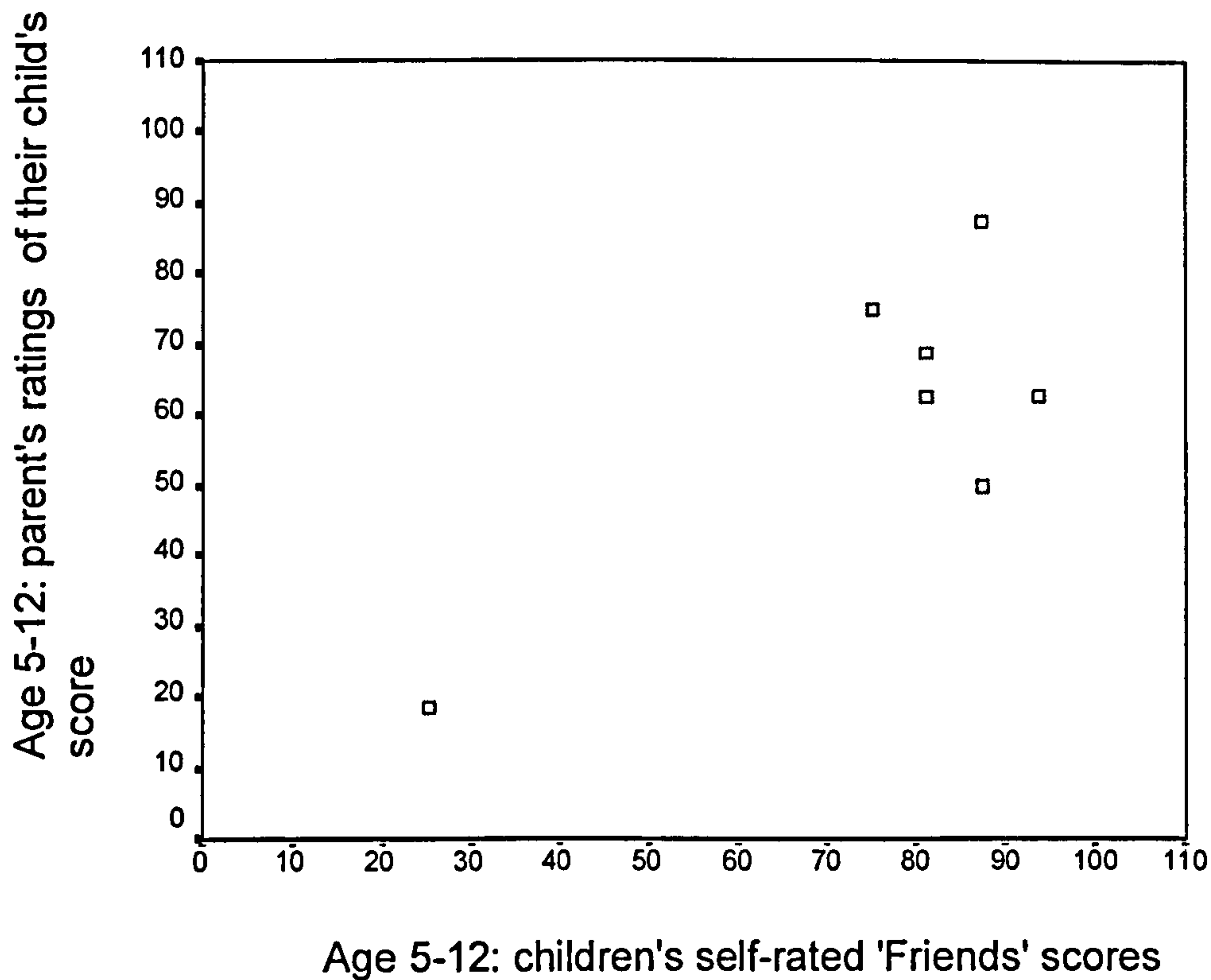
Although not the primary focus of the analysis, when scores were analysed according to gender, 5 boys had significantly higher Total Quality of Life scores ($P=0.02$, CI 1.75-19.35), and significantly higher scores on the domains of Friends ($P=0.001$; CI 23.03 – 40.72) and Disease ($P=0.01$; CI 7.84- 42.15) than 3 girls. These data provide some evidence that girls and boys make different judgements when scoring domains; these findings however need to be replicated in larger studies.

Ventilator-dependent children aged 5-12 compared with parents

Although Total Quality of Life scores failed to reach significance, 17 parents had significantly lower scores than 8 ventilator-dependent children on 3 domains: Emotional wellbeing ($P=0.02$); Friends ($P=0.04$); and Disease ($P=0.01$). Friends was the only domain that had a positive correlation between scores ($r=0.78$, $P=0.03$).

Findings from the 8 matched pairs of children and parents only reached significance on the domain of Friends ($P=0.03$), which also had a positive correlation between scores ($r=0.78$, $P=0.03$) (Table 3.1). The association between scores is illustrated in Graph 1. There were also gender differences that did not reach statistical significance but are worthy of note. Parents of 5 boys scored lower aggregated mean scores across all domains than their sons (apart from School), whereas parents of 3 girls scored higher aggregated mean across all domains (apart from Emotional wellbeing and School) than their daughters. These gender differences need to be interpreted with caution as numbers are low and aggregated mean scores can mask the range of scores, but nonetheless these data indicate that parents make some differentiation according to their child's gender.

Graph 3.1. Scatterplot of matched pairs of parents and children aged 5-12 years for the KINDL domain of 'Friends'.



KINDL: Children aged 13-19 years

Data were obtained from 9 ventilator-dependent children and 11 parents, and scores were compared with normative reference values from 583 school children. Findings are summarised in Table 3.2.

As predicted, Total Quality of Life scores for ventilator-dependent children aged 13-19 years were significantly lower than for school children ($P=0.009$).

In addition, teenage ventilator-dependent children had significantly lower Self-esteem ($P= 0.01$) than school children of a similar age. Self-esteem was not an issue with younger ventilator-dependent children and this finding adds weight to the argument that it is important to consider children and teenagers separately.

Gender differences: Children aged 13-19 years

Within this older age group there was no significant difference in Total Quality of Life scores between boys and girls. Teenage boys had the lowest mean Self-esteem scores across all groups of children but the difference just failed to reach significance when compared with teenage girls.

Ventilator-dependent children aged 13-19 compared with parents

As with younger children, Total Quality of Life scores failed to show a significant difference, but in contrast there was a strong positive correlation between scores ($r=0.84$, $P=0.004$) (Table 3.2). Eleven parents had significantly lower scores than 9 teenage children on 2 domains: Physical wellbeing ($P=0.05$), and Disease ($P=0.02$). However, teenage children reported significantly lower Self-esteem than parents ($P=0.005$). Only Self-esteem and School did not show a positive correlation between teenage children and parents.

Findings from the 9 matched pairs of teenage children and their parents indicated that parents had significantly lower scores on the domains of Physical wellbeing ($P=0.01$), Self-esteem ($P=0.05$), and Disease ($P=0.004$) (Table 3.2). All domains showed a positive correlation apart from School. The gender differences that were found with parents of younger children were not so pronounced amongst parents and their teenage children. Total Quality of Life scores of 6 parents and their teenage sons were positively correlated but not significantly different. Parents of adolescent boys scored significantly lower on the domains of Physical wellbeing ($P=0.03$, CI -19.9 to -7.09; $r=0.88$, $P=0.02$) and Disease ($P=0.03$, CI -34.1 to -2.0) than their sons and significantly higher on Self-esteem ($P=0.01$, CI 10.3 - 48.0). Whereas there were no significant differences between 3 parents and their adolescent daughters.

Table 3.2. Summary of KINDL results: Children aged 13 – 19 years. Original in colour.

	Normative Reference values School Children aged over 13 years N=583						Ventilator-dependent children Aged 13 – 19 years N= 9						Parents of ventilator-dependent children Aged 13-19 years N=11						Mean differences: Ventilator-dependent children compared with School children	Mean differences: 11 Parents compared with 9 ventilator-dependent children	Mean differences: 9 parents matched with their 9 ventilator- dependent children									
	Boys			Girls			Total			Boys (n=6)			Girls n=3			Total						Boys (n=7)			Girls (n=4)			Total		
	Mean	SD		Mean	SD		Mean	SD		Mean	SD		Mean	SD		Mean	SD					Mean	SD		Mean	SD		Mean	SD	
Total Quality of Life Score	73.53	8.83		70.78	10.01		72.15		58.81	13.25		59.30	9.14		58.98		61.97	17.90	61.97	15.31		61.74		Ventilator-dependent children had significantly lower scores than school children P=0.009 (-21.95 to -4.38)	Correlation r=0.84 P=0.004	Correlation r=0.84 P=0.004				
Physical wellbeing	77.18	13.07		68.24	17.38		72.71		67.70	12.75		70.83	20.09		68.75		54.16	10.20	61.25	23.96		57.38		NS	Correlation r=0.82 P=0.006 Parents had significantly lower scores than children P=0.05 (-22.91 to - 0.18)	Correlation r=0.82 P=0.006 Parents had significantly lower scores than their children: P=0.001 (-21.6 to -8.87)				
Emotional wellbeing	79.49	11.80		79.41	12.89		79.45		70.83	8.53		66.66	23.66		69.44		67.70	16.49	61.25	19.96		64.77		NS	Correlation r=0.87 P=0.002	Correlation r=0.87 P=0.002				
Self-esteem	63.27	19.34		58.14	19.06		60.70		26.04	19.53		45.83	34.42		32.63		55.20	24.81	60.00	23.63		57.38		Ventilator-dependent children had significantly lower scores than school children P= 0.01 (-47.36 to -8.69)	Parents had significantly higher scores than children: P=0.005 (9.17 to 40.33)	Children had significantly lower scores than their parents: P=0.05 (0.22 to 51.61)				
Family	79.56	17.05		75.51	17.68		77.53		68.75	17.67		52.08	23.66		63.19		70.83	16.13	62.50	11.69		67.04		NS	Correlation r=0.81 P=0.007	Correlation r=0.81 P=0.007				
Friends	78.43	11.96		78.06	13.47		78.24		58.33	38.45		68.75	12.50		61.80		58.33	33.46	60.00	14.38		59.09		NS	Correlation r=0.90 P=0.001	Correlation r=0.90 P=0.001				
School	63.58	14.04		65.19	13.21		64.38		57.50	17.34		53.12	22.09		56.25		65.62	24.92	70.31	13.85		67.50		NS	NS	NS				
Disease *							78.67		79.86	23.77		80.00	18.02		79.90		61.80	22.42	66.66	17.17		64.01		NS	Correlation 0.81 P=0.007 Parents had significantly lower scores than children: P=0.02 (-28.91to - 2.85)	Correlation r=0.81 P=0.007 Parents had significantly lower scores than their children: P=0.004 (-26.1to - 6.78)				

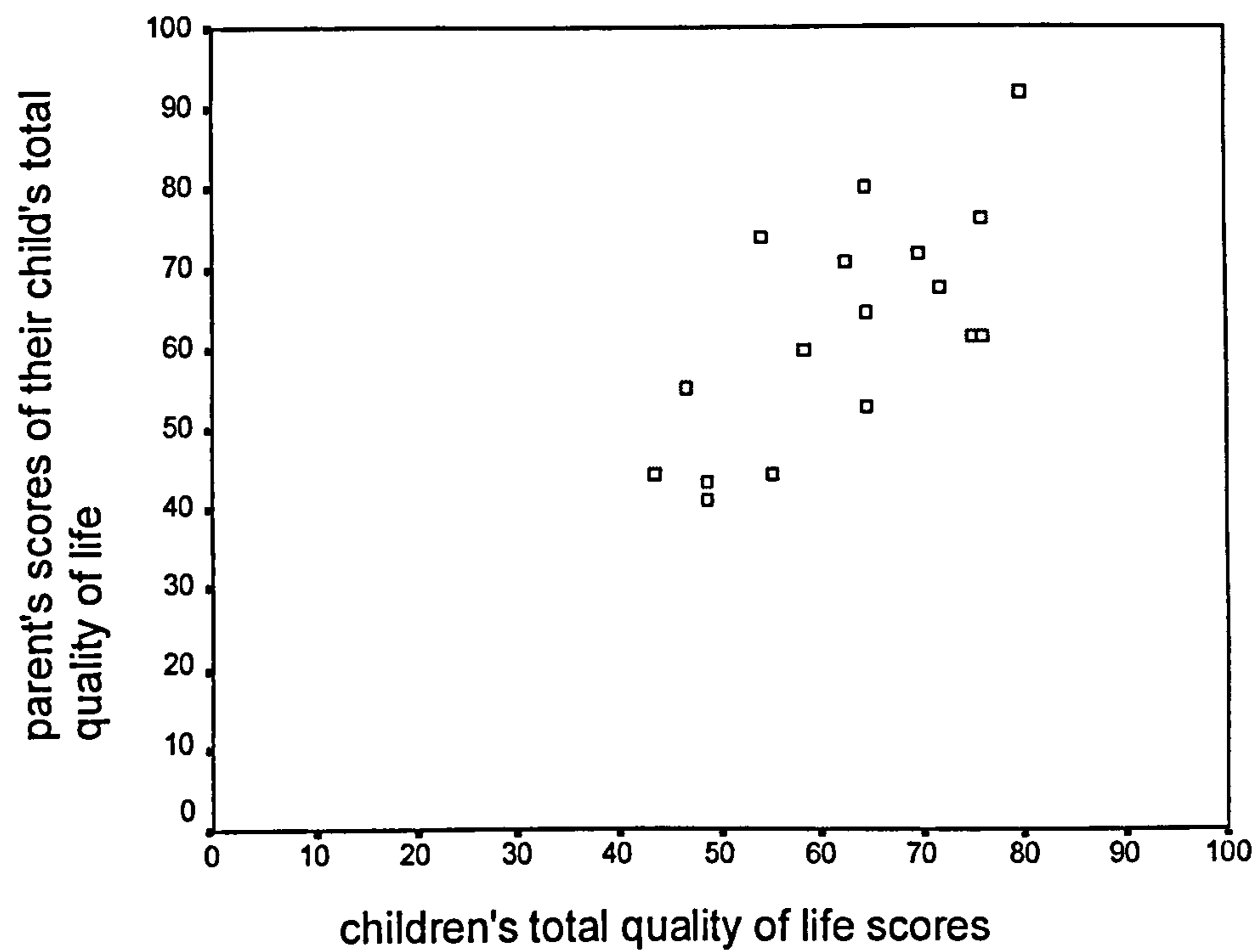
* Derived from a population of 1050 chronically ill children

KINDL: All ventilator-dependent children aged 5-19, compared with school children and parents

Combined data from 17 children aged 5-19 years and 28 parents were compared with 1501 school children and Disease scores of 1050 chronically ill children. Findings are summarised in Table 3.3. When the two subgroups were combined into one group, children dependent on ventilators reported significantly lower Total Quality of Life scores ($P=0.0009$), and significantly lower scores on all domains except Friends compared with school children, and chronically ill children in respect of Disease. Ravens-Sieberer and Bullinger (2000) subsequently provided mean scores derived from groups of children with asthma, atopic dermatitis and obesity. A temporal perspective was observed both on a sub-scale level and with Total Quality of Life scores; ventilator-dependent children had lower scores across every sub-scale and across Total Quality of Life scores compared with all other groups of chronically ill children. For example, Total Quality of Life scores across groups were as follows: atopic dermatitis 74.41, asthma 73.38, obesity 68.93, and ventilator-dependent children 62.40.

Although no significant difference was found in Total Quality of Life scores, parents reported significantly lower scores on 2 domains (Emotional wellbeing $P=0.03$ and Disease $P=0.007$). However, children scored significantly lower scores than parents on the Self-esteem domain ($P=0.002$). All scores apart from Self-esteem and School had a positive correlation. For example, Total Quality of Life scores had a correlation coefficient of $r=0.78$ ($P=0.03$) and are illustrated in a Scatterplot (Graph 3.2)

Graph 3.2. Scatterplot of 17 matched pairs of parents and children aged 5-19 years for the KINDL Total Quality of Life Scores.



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Table 3.3. Summary of KINDL results: All children aged 5-19 years. Original in colour.

	Normative reference values N=1501 School children Mean scores	All children dependent on ventilators N=17 Age 5-19 Mean scores	All parents of children dependent on ventilators N=28 Age 5-19 Mean scores	All ventilator-dependent children compared with normative children N=17 Aged 5-19 years	All parents compared with all ventilator-dependent children N=17 children aged 5-19 years and 28 parents	All 17 ventilator-dependent children compared with their matched parents
Total Quality of Life Score	76.75	62.40	63.71	Ventilator-dependent children scored significantly lower than normative children P=0.0009 (-20.23 to -8.46)	Correlation r=0.70 P=0.001	Correlation r=0.70 P=0.001
Physical wellbeing	75.56	63.28	58.79	Ventilator-dependent children scored significantly lower than normative children P=0.004 (-19.86 to -4.86)	Correlation r=0.53 P=0.03	Correlation r=0.53 P=0.03
Emotional wellbeing	83.01	73.04	65.74	Ventilator-dependent children scored significantly lower than normative children P=0.014 (-17.62 to -2.29)	Correlation r=0.52 P=0.03 Parents scored significantly lower than children P=0.03 (-14.13 to -0.46)	Correlation r=0.52 P=0.03
Self-esteem	66.60	45.31	60.18	Ventilator-dependent children scored significantly lower than normative children P=0.005 (-35.12 to -7.44)	Parents scored significantly higher than children P=0.002 (5.90 to 23.84)	NS
Family	83.99	67.96	70.37	Ventilator-dependent children scored significantly lower than normative children P=0.006 (-26.61 to -5.4)	Correlation r=0.78 P=0.001	Correlation r=0.78 P=0.001
Friends	78.18	67.96	63.42	NS	Correlation r=0.84 P=0.00004	Correlation r=0.84 P=0.00004 Parents had significantly lower scores than children P=0.04 (-16.24 to -0.16)
School	73.13	60.09	66.92	Ventilator-dependent children scored significantly lower than normative children P=0.008 (-21.96 to -4.1)	NS	NS
Disease *	78.67	79.06	62.80	NS	Correlation r=0.51 P=0.03 Parents scored significantly lower than children P=0.007 (-24.40 to -8.10)	Correlation r=0.51 P=0.03 Parents had significantly lower scores than children P=0.007 (-27.48 to -5.16)

*Derived from a population of 1050 chronically ill children

Findings from the 17 matched pairs of ventilator-dependent children and their parents indicated that parents' Total Quality of Life scores were not significantly different, but parents' scores were significantly lower on the domains of Friends ($P=0.04$), and Disease ($P=0.007$) (Table 3.2). All domains had a positive correlation apart from School and Self-esteem. For example, on the domain of Friends, scores had a correlation coefficient of $r=0.84$ ($P=0.0004$).

On an individual level, there were subtle differences in scores between matched pairs of parents and children. Two examples of parent's scores matched with their children's scores are illustrated in Table 3.4. In particular, both parents had significantly higher Self-esteem scores than their children. These findings add weight to the different perceptions and judgements between parents and their children

Table 3.4. Two examples of individual differences in scores attributed by parents and their children.

	Case X Male aged 13		Case Y Female aged 15	
	Child	Parent	Child	Parent
KINDL				
Total Quality of Life	41.67	44.79	68.75	80.21
Physical wellbeing	50.00	43.75	93.75	75.00
Emotional wellbeing	68.75	56.25	93.75	87.50
Self-Esteem	0.00	43.75	12.50	100.00
Family	68.75	56.25	68.75	56.25
Friends	0.00	0.00	81.25	81.25
School	62.50	68.75	62.50	81.25
Disease	37.50	33.33	100	87.50
EQ-5D				
Mobility	3	3	3	3
Self care	3	3	1	1
Usual activities	2	3	1	2
Pain/discomfort	1	2	1	1
Anxiety/depression	1	2	1	1
Health	30	33	100	100
Quality of life	40	33	100	90
Health Index	0.09	-0.17	0.34	0.30

EQ-5D

Findings were compared with normative data obtained from a survey undertaken with over 3,000 children attending primary and secondary schools in England.

Health dimensions

There were insufficient numbers of ventilator-dependent children in each age band (5-12 and 13-19 years) to undertake subgroup analyses using the chi square statistic for each of the 5 health dimensions as too many categories contained no ventilator-dependent children. Therefore results are reported for the entire group and illustrated in Table 3.5.

Significantly more moderate or severe problems ($P=0.0000$) were reported by all 17 ventilator-dependent children and 28 parents compared to school children on all dimensions except for anxiety/depression (Table 3.5). For example, in relation to mobility, 71% of ventilator-dependent children reported a problem, whereas with school children the percentage was 4.6%. In relation to the domain of anxiety/depression, 12% (2/17) of ventilator-dependent children reported problems, whereas in school children the figure was 28%. In contrast, 61% of parents reported significantly more moderate and severe problems in their ventilator-dependent children.

Table 3.5. Numbers and percentages of school children, all ventilator-dependent children (aged 5-19) and parents reporting a moderate or severe problem in each EQ-5D dimension.

Problem				Chi Square
Euroquol dimension	Moderate	Severe	Any	
Mobility				
School children	139/3130 4.4%	5/3130 0.2%	144/3130 4.6%	Ventilator-dependent children and parents report significantly more problems (especially severe) than school children. P=0.0000
All Ventilator-dependent children	2/17 11.8%	10/17 58.8%	12/17 70.6%	
Parents of all ventilator-dependent children	2/28 7.1%	20/28 71.4%	22/28 78.5%	
Self care				
School children	30/3127 1%	6/3127 0.2%	36/3127 1.2%	Ventilator-dependent children and parents report significantly more problems (especially severe) than school children. P=0.0000
All Ventilator-dependent children	1/17 5.9%	10/17 58.8%	11/17 64.7%	
Parents of ventilator-dependent children	6/28 21.4%	16/28 57.1%	22/28 78.5%	
Usual activities				
School children	225/3127 7.2%	4/3127 0.1%	229/3127 7.3%	Ventilator-dependent children and parents report significantly more problems (especially severe) than school children. P=0.0000
All Ventilator-dependent children	7/17 41.2%	1/17 5.9%	8/17 47%	
Parents of ventilator-dependent children	12/28 42.9%	8/28 28.6%	20/28 71.4%	
Pain/discomfort				
School children	731/3126 23.4%	32/3126 1%	763/3126 24.4%	Ventilator-dependent children and parents report significantly more moderate problems than school children. P=0.0000 Parents reported more problems than ventilator-dependent children. P=0.0000
All Ventilator-dependent children	6/17 35.3%	0/17 0%	6/17 35.3%	
Parents of ventilator-dependent children	20/28 71.4%	0/28 0%	20/28 71.4%	
Anxiety/depression				
School children	787/3123 25.2%	78/3123 2.5%	865/3123 27.7%	Ventilator-dependent children have significantly less moderate problems and no severe problems than school children. P=0.0000. Parents report significantly more moderate and severe problems in their ventilator dependent children than school children. P=0.0000
All Ventilator-dependent children	2/17 11.8%	0/17 0%	2/17 11.8%	
Parents of ventilator-dependent children	15/28 53.6%	2/28 7.1%	17/28 60.7%	

On an individual level, as with the KINDL, there were subtle differences in scores between individual pairs of parents and children. Two examples were illustrated previously in Table 3.4. In particular, the parent of child X in Table 3.4, reported that her child had moderate problems on the domains of pain/discomfort and anxiety/depression whereas her son reported that he had no problems.

Health state

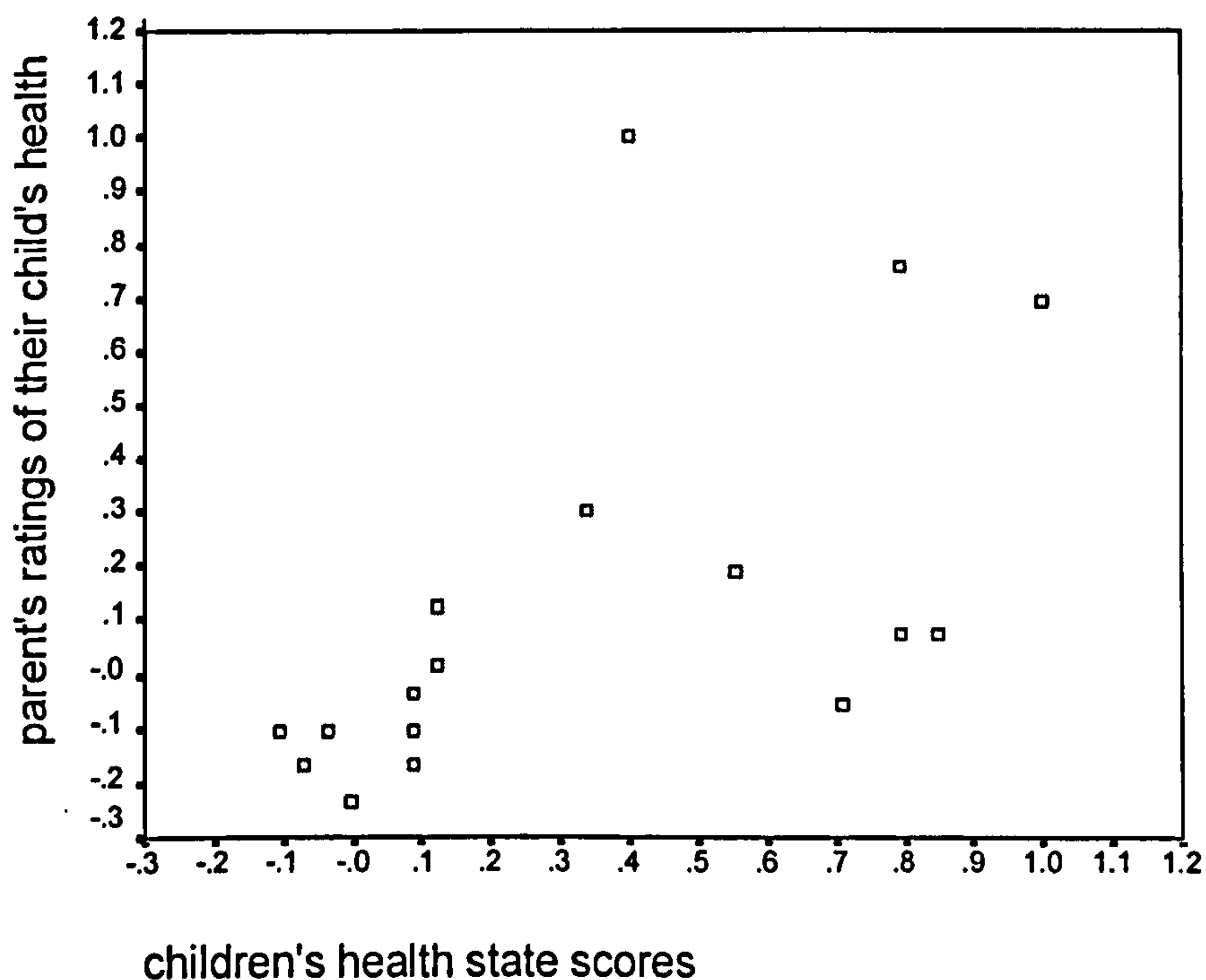
The mean Health State score for ventilator-dependent children was 0.34 (SD 0.40) (Table 3.6). In contrast to the gender differences identified with the KINDL, there were no significant within-group differences between ventilator-dependent boys, girls or age group. Ventilator-dependent children (boys and girls and both age groups) had significantly lower scores than school children ($P=0.00001$). This may be explained by the fact that health and impairment as identified by the EQ-5D was similar across age groups and genders, whereas children of different age groups and gender made different judgements concerning the emotion-focused domains that contribute to the overall KINDL Total Quality of Life score. A similar phenomenon was seen in the test cases whereby people with a similar level of ventilator-dependency and impairment made different judgements concerning their quality of life.

In contrast to the KINDL, parents had significantly lower scores than ventilator-dependent children ($P=0.02$). There was a weak correlation between the Health State scores of parents and children ($r=0.58$, $P=0.01$), which is illustrated in Graph 3.3.

Table 3.6. Health State scores: EQ-5D.

Age group	Children Aged 4-12	Children Aged 13-19	Total	Outcome t- test
Health State				
School children	0.90 1416	0.87 1679	Mean 0.89 N 3095 SD 0.16	
Boys	0.90 712	0.90 802	Mean 0.90 N 1514 SD 0.16	
Girls	0.90 704	0.86 877	Mean 0.88 N 1581 SD 0.15	
Ventilator-dependent children	0.36 8 0.38	0.31 9 0.38	Mean 0.34 N 17 SD 0.40	Ventilator-dependent children (boys and girls and both age groups) had significantly lower scores than school children P=0.00001 (-0.74 to -0.36)
Boys	0.37 5 0.48	0.30 6 0.41	Mean 0.33 N 11 SD 0.42	
Girls	0.35 3 0.23	0.32 3 0.39	Mean 0.34 N 6 SD 0.29	
Parents	0.24 17 0.45	0.01 11 0.25	Mean 0.16 N 28 SD 0.39	Parents had significantly lower scores than their ventilator-dependent children P=0.02 (-0.32 to -0.02)
Boys	0.22 11 0.43	0.03 9 0.28	Mean 0.13 N 17 SD 0.40	
Girls	0.22 6 0.48	-0.07 2 0.02	Mean 0.21 N 11 SD 0.40	
				Correlation r=0.58 P=0.01

Graph 3.3. Scatterplot of 17 matched children's and parent's self-reported Health State scores on the EQ-5D.



Self-rating of health reported on the visual analogue scale

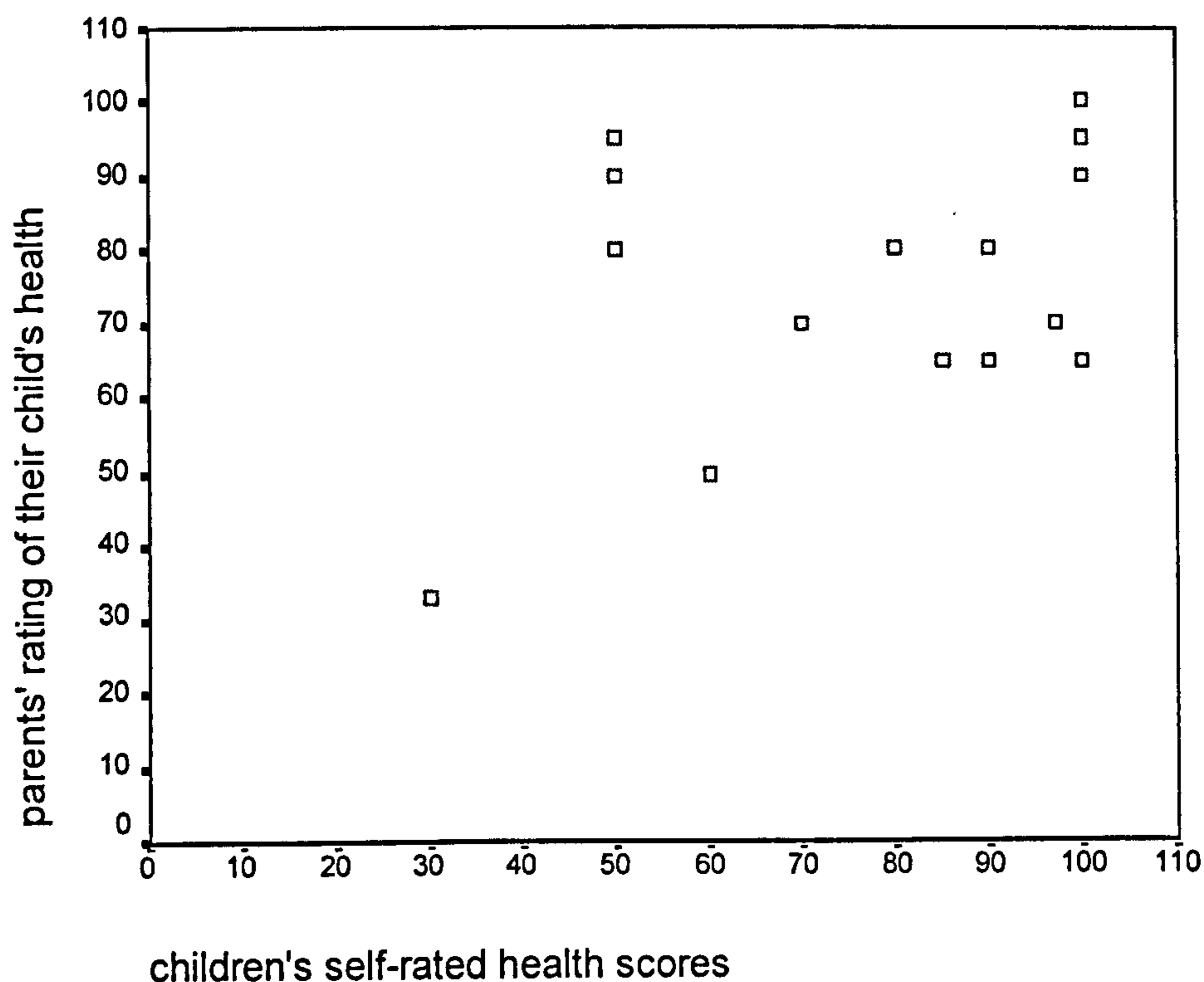
The mean self-rating of health for ventilator-dependent children of all age groups was 78.35 (SD 22.59) (Table 3.7). There were no significant within-group differences between boys, girls or age group; nor was there a significant difference between ventilator-dependent children and school children, or parents and ventilator-dependent children.

Table 3.7. Self-rating of health reported on the visual analogue scale EQ-5D.

Age group	Children Aged 4-12	Children Aged 13-19	Total	Outcome t-test
Visual Analogue Scale Self-Rating of Health				
School children	89.97 1410	76.48 1668	Mean 83.17 N 3078 SD 15.59	
Boys	88 701	81 891	Mean 84.26 N 1497 SD 15.47	
Girls	90 709	74 872	Mean 82.15 N 1581 SD 15.64	
Ventilator-dependent children	84.62 8 22.41	72.78 9 22.51	Mean 78.35 N 17 SD 22.59	There was no significant difference between ventilator-dependent children (boys or girls, or between age groups) and school children
Boys	89.40 5 22.06	70.83 6 24.98	Mean 79.27 N 11 SD 24.51	
Girls	76.67 3 25.16	76.67 3 20.81	Mean 76.67 N 6 SD 20.65	
Parents	80.00 17 15.38	71.64 11 19.27	Mean 78.39 N 28 SD 17.56	There was no significant difference between parents and ventilator-dependent children
Boys	75.00 11 16.03	68.83 6 20.05	Mean 74.71 N 17 SD 17.50	
Girls	95.00 6 9.82	75.00 5 20.00	Mean 84.09 N 11 SD 16.85	

Amongst the 17 matched pairs of ventilator-dependent children and their parents, two thirds of children ascribed a different score to their self-rated health than their parents: 8 children recorded a higher and 4 a lower score than their parents (Graph 3.4). The scores were not correlated or statistically different, but findings indicate that, as with the KINDL, children and their parents do make different judgements about health.

Graph 3.4. Scatterplot of 17 matched children's and parent's self-reported health scores with the EQ-5D.



In addition, ventilator-dependent children and parents were asked to compare the child's health with that of other children of the same age. Of the 15 children who responded: 8 reported their health as the same; 7 said their health was worse or much worse. In contrast, of the 28 parents who responded: 2 said that their child's health was better or much better, a quarter said the same and just over two thirds said their child's health was worse or much worse.

Self-rating of quality of life reported on the visual analogue scale

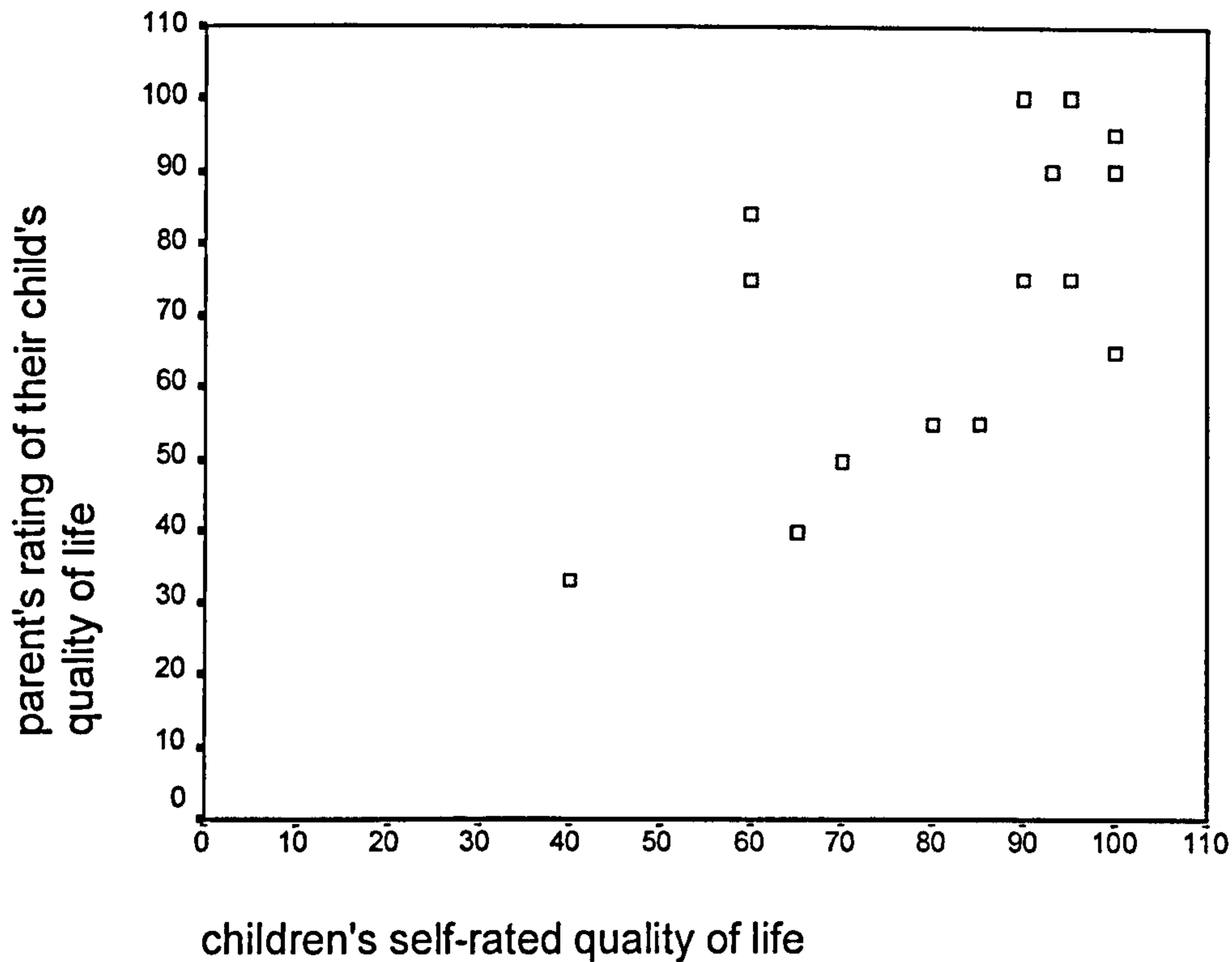
The mean self-rating of quality of life for ventilator-dependent children of all age groups was 83.71 (SD 18.26) (Table 3.8). There were no significant within-group differences between boys, girls or age group. There are no normative quality of life reference values available for school children for comparison. Parents as a whole group had significantly lower quality of life scores than ventilator-dependent children ($P=0.02$).

Table 3.8. Self-rating of quality of life reported on the visual analogue scale EQ-5D.

Age group	Children Aged 4-12	Children Aged 13-19	Total	Outcome of t-test
Visual Analogue Scale Self-Rating of Quality of life				
Ventilator-dependent children	90.38 8 14.43	77.78 9 20.01	Mean 83.71 N 17 SD 18.261	Parents had significantly lower scores than ventilator-dependent children P=0.02 (-17.71 to -1.64) Correlation r=0.67 P=0.003
Boys	97.60 5 3.62	75.83 6 21.54	Mean 85.73 N 11 SD 19.126	
Girls	78.33 3 18.93	81.67 3 20.20	Mean 80.00 N 6 SD 17.607	
Parents	75.65 17 22.48	69.27 11 22.21	Mean 74.04 N 28 SD 20.71	
Boys	75.55 11 22.50	65.27 9 23.36	Mean 73.18 N 17 SD 22.083	
Girls	75.83 6 24.58	84.50 2 0.70	Mean 75.36 N 11 SD 19.377	

Amongst the 17 matched pairs of ventilator-dependent children and their parents, all children ascribed a different score to their self-rated quality of life than their parents: 14 children recorded a higher and 3 a lower score than their parents. (Graph 3.5). The matched pairs of quality of life scores had a positive correlation ($r=0.67$, $P=0.003$) and were significantly different with parents recording significantly lower scores than their children ($P=0.02$).

Graph 3.5. Scatterplot of 17 matched children's and parent's self-reported quality of life scores on the EQ-5D.



In addition, ventilator-dependent children and their parents were asked to compare the child's quality of life with that of other children of the same age (Table 3.9). Of the 15 children who responded: a quarter reported their health as better or much better; around half said the same; and a further quarter said it was worse. In contrast, of the 28 parents who responded: around a quarter said that their child's health was better or much better, a quarter said the same and just under half said their child's health was worse or much worse.

Health during the last 12 months

Parents reported significantly fewer ventilator-dependent children with very good health and more ventilator-dependent children with poor health than school children (P=0.001) (Table 3.9).

Table 3.9. Judgements about health and quality of life during the last 12 months EQ-5D.

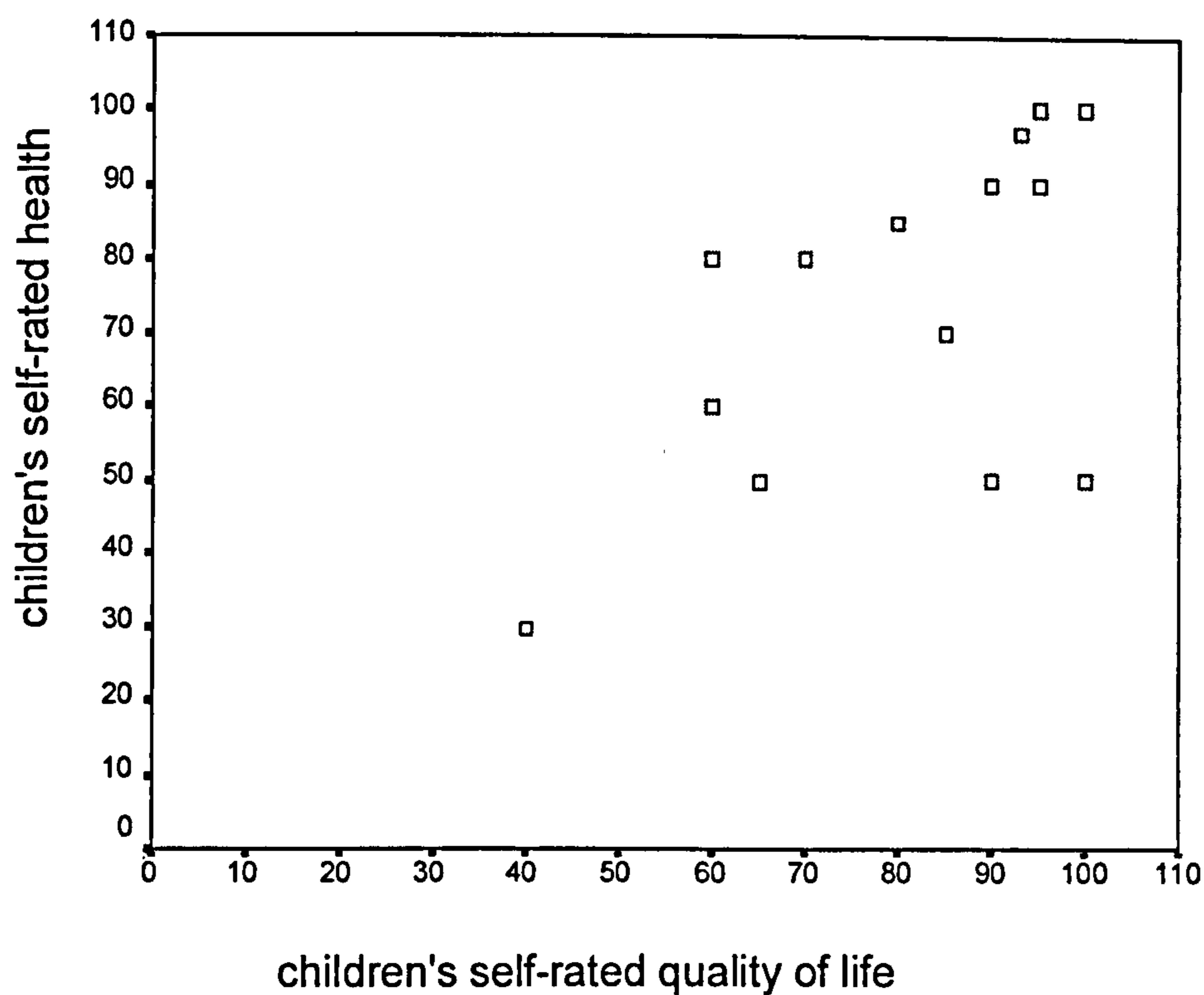
Health in comparison to other children						
Euroquol	Much better	Better	Same	Worse	Much Worse	Chi Square
Ventilator-dependent children	0/15 0%	0/15 0%	8/15 53%	6/15 40%	1/15 6.6%	NS
Parents of ventilator-dependent children	1/28 3.5%	1/28 3.5%	7/28 25%	11/28 39%	8/28 28%	
Quality of life in comparison to other children						
	Much better	Better	Same	Worse	Much Worse	
Ventilator-dependent children	2/15 11.7%	2/15 11.7%	7/15 46%	4/15 26%	0/15 0%	NS
Parents of ventilator-dependent children	3/28 10.7%	3/28 10.7%	7/28 25%	10/28 25%	5/28 17.8%	
Self-rated health during last 12 months						
	Very good	Good	Fair	Poor	Very poor	
School children	815/3095 26%	1705/3095 55%	496/3095 16%	71/3095 2.3%	8/3095 0.25%	Parents reported significantly fewer ventilator-dependent children with very good health and more ventilator-dependent children with poor health than school children. P=0.001
Parents of ventilator-dependent children	4/35 11.4%	18/35 51.4%	9/35 25.7%	4/35 11.4%	0/35 0%	

Comparison of health and quality of life

Children

Around two thirds of children ascribed a different score to their self-rated health and quality of life; 5 children reported that their health was higher than their quality of life and 6 said it was lower (Graph 3.6). Although the scores were positively correlated ($r=0.63$, $P=0.004$) and were not significantly different, findings indicate that children do make some differentiation between their health and quality of life when measured on a visual analogue scale.

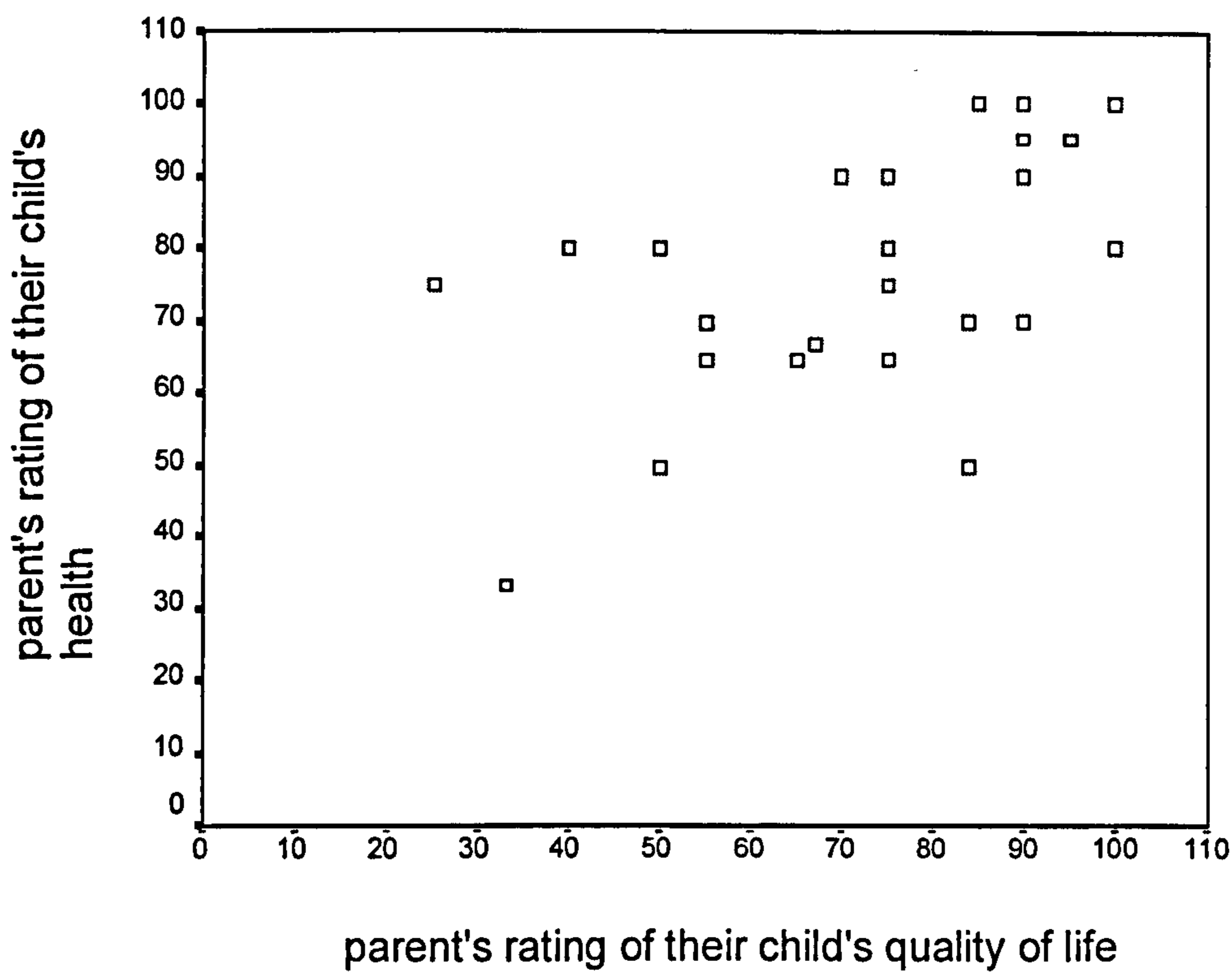
Graph 3.6. Scatterplot of children's self-reported health and quality of life scores with the EQ-5D.



Parents

Similar to children, around two thirds of parents ascribed a different score to the self-rated assessment of their child's health and quality of life (Graph 3.7). Thirteen parents assessed that their child's health was higher than their quality of life and 6 parents reported it was lower. Although the scores were positively correlated ($r=0.59$, $P=0.001$) and were not statistically different, findings indicate that parents also make some differentiation between their children's health and quality of life.

Graph 3.7. Scatterplot of parent's self-reported health and quality of life scores in relation to their children (EQ-5D).



Health-related quality of life of ventilator-dependent children living in hospital compared with ventilator-dependent children living at home

The scores of 2 children and proxy-ratings of 4 parents of children living in hospital were compared with scores of 15 children and the proxy-ratings of parents of 23 children living at home (Table 3.10). Results need to be interpreted with caution as numbers were low.

2 ventilator-dependent children living in hospital compared with 15 children living at home

Although Total Quality of Life scores on the KINDL were not significantly different, children at home had significantly higher scores on 4 domains (Physical wellbeing (P=0.01); Self-esteem (P=0.02); Friends (P=0.0002); and Disease (P=0.005). On the EQ-5D, children at home had significantly higher overall Health State scores (P=0.01), health (P=0.001) and quality of life (P=0.00000) scores than children in hospital.

4 parents of ventilator-dependent children living in hospital compared with 23 parents of ventilator-dependent children living at home

Total Quality of Life scores on the KINDL were significantly higher ($P=0.005$) for children living at home but overall Health State scores on the EQ-5D were not significantly different.

Table 3.10. Health-related quality of life of children in hospital compared with children living at home. Original in colour.

KINDL	Children in hospital N=2	Children at home N=15	Parents of children in hospital N=4	Parents of children at home N=23	Children at home compared with children in hospital	Parents of children at home compared with parents of children in hospital
Total Quality of Life Score	59.8	62.73	56.58	64.95	NS	Children at home had significantly higher scores P=0.005 (2.73 to 14.00)
Physical wellbeing	53.1	64.73	53.12	59.78	Children at home had significantly higher scores P=0.01 (3.18 to 20.07)	NS
Emotional wellbeing	78.1	72.32	54.68	67.66	NS	Children at home had significantly higher scores P=0.001 (6.13 to 19.83)
Self-Esteem	31.2	47.32	51.56	61.68	Children at home had significantly higher scores P=0.02 (2.00 to 30.13)	Children at home had significantly higher scores P=0.03 (1.03 to 19.21)
Family	84.3	65.62	71.87	70.10	Children at home had significantly lower scores P=0.03 (-29.8 to -7.6)	NS
Friends	40.6	71.87	51.56	65.48	Children at home had significantly higher scores P=0.0002 (17.94 to 44.60)	Children at home had significantly higher scores P=0.001 (6.42 to 21.43)
School	71.8	57.95	71.87	65.93	Children at home had significantly lower scores P=0.01 (-23.9 to -3.71)	NS
Disease	62.5	81.27	53.12	64.49	Children at home had significantly higher scores P=0.005 (6.65 to 30.90)	Children at home had significantly higher scores P=0.006 (3.65 to 19.09)
EQ-5D						
VAS Health	55.0	81.47	72	79.46	Children at home had significantly higher scores P=0.001 (15.32 to 37.62)	Children at home had significantly higher scores P=0.03 (0.77 to 14.14)
VAS Quality of life	55.0	87.53	48.25	78.33	Children at home had significantly higher scores P=0.00000 (24.40 to 40.66)	Children at home had significantly higher scores P=0.00000 (23.06 to 37.10)
Health State	0.09	0.37	0.17	0.22	Children at home had significantly higher scores P=0.01 (0.06 to 0.49)	NS

3.5 Discussion

Taking the group of 17 ventilator-dependent children as a whole, the hypothesis that ventilator-dependent children would have significantly lower health-related quality of life than school children was supported on two fronts: Findings derived from the KINDL confirmed that ventilator-dependent children had significantly lower quality of life and findings derived from the EQ-5D confirmed that ventilator-dependent children had significantly lower health states than school children. KINDL scores also demonstrated that the instrument was able to discriminate between differences in the impairment of health-related quality of life compared with other groups of chronically ill children. A temporal association was observed both on a sub-scale level and in Total Quality of Life scores compared with 3 groups of chronically ill children, with ventilator-dependent children having the lowest observed scores. Compared to the other 3 groups of chronically ill children with asthma, atopic dermatitis and obesity, ventilator-dependent children are likely to have more health-related problems due to the severity of their underlying disease and co-morbidity.

As a group, ventilator-dependent children were found to have significantly lower self-esteem than school children. This finding suggests that the KINDL may be useful in predicting or classifying children who would benefit from a therapeutic intervention aimed at raising their self-esteem. Findings from the KINDL also add weight to the impact of gender and a child's development on their perception and decision-making processes concerning their health and quality of life.

Nonetheless, the EQ-5D did throw up some anomalies in that ventilator-dependent children's visual analogue scale scores concerning their health on the day of data collection were not significantly lower than a general population of school children. This outcome appeared surprising to members of the EuroQol team advising the study; they felt it might have been expected that children with such profound physical impairments and obvious health problems would rate their health lower than they did. This could be explained by the fact that the EQ-5D represents a utility measurement whereby responses given to

the 5 questions of the measure are weighted by values derived from a representative population of predominately non-disabled school children (Jenkinson *et al.* 1997). It may be that for ventilator-dependent children, many of who have profound physical impairments, that independent mobility and being able to care for themselves may be less important than the ability to enjoy life generally. These issues will be explored when qualitative and quantitative findings around health-related quality of life are synthesised in Chapter 4.

In addition, when parents and children were asked to make a judgement as to how their health and quality of life compared with other school children of the same age, half of the children and two thirds of parents said that the health of their children was the same or better than other children. Around three quarters of children and half of the parents said that their quality of life was either the same or better than other children. These findings appear to contradict the overall low ratings of health-related quality of life derived from the KINDL and EQ-5D and warrant further investigation.

The children and young people measured their health on a visual analogue state from 1-100 (with 1 being the worst imaginable health and 100 being the best imaginable health). There was no significant difference found between the estimates of health by ventilator-dependent children and a normative population of school children. This may indicate that the visual analogue scale represents a more appropriate assessment as to how ventilator-dependent children view their health, which is not perceived as being very different to other predominantly non-disabled children.

The EQ-5D predominantly measured functional status and for the children and young people children who had severe physical impairments there was a constant floor effect. It is difficult to see how this subgroup of children who are dependent on ventilators could improve their state of health when measured with the EQ-5D when they are unlikely, for example, to be able to regain physical functioning following a paralysing head injury. Some of the children also have life-limiting conditions (such as muscular dystrophy) which means that their health will continue to deteriorate steadily until death. Given that the

EQ-5D is one measure on which calculations for resource allocation and prioritisation might be made, it is difficult to interpret results when this group of ventilator-dependent children recorded a significantly lower health state and yet reported that their health was the same as other predominantly non-disabled school children on the visual analogue scale. Nonetheless, as one might expect in terms of a temporal relationship, findings from the EQ-5D indicate that ventilator-dependent children have poor (if not very poor) health states relative to a general population of school children.

Most of the group of ventilator-dependent children would be dead or have severe life-limiting respiratory distress without the ventilator. Therefore it could be argued that the addition of a ventilator has lifted children from a worst possible state of health (-0.59 death) to a baseline mean utility weighted score of 0.34 compared with 0.89 for the general population of school children.

The second hypothesis was that parents would report significantly lower health-related quality of life than their ventilator-children. Here a more complex picture emerged making interpretation difficult and less clear-cut. In terms of health-related quality of life, the hypothesis could not be supported conclusively. Overall with the KINDL, when comparing all 17 children with all 28 parents, no significant difference was found in Total Quality of Life scores, although parents reported significantly lower scores on 2 domains (Emotional wellbeing $P=0.03$ and Disease $P=0.007$). However, children scored significantly higher scores than parents on the Self-esteem domain ($P=0.002$). Although numbers were small, there were differences in outcome between age bands of children and parents and matched pairs of children and parents. For example, in the aged 5-12 year band, 8 matched pairs of children and their parents only reached a significant difference on the domain of Friends, whereas when the same 8 children were compared with all 17 parents of children aged 5-12 years, parents assigned significantly lower scores than children in 3 domains (Emotional wellbeing; Friends and Disease). This may reflect the fact that some children who did not complete questionnaires were unable to do so because they had profound sensory impairments and their parent's proxy estimates of these profoundly disabled children may have skewed the results.

Findings from the EQ-5D add weight to findings from the KINDL that parents report significantly more 'Disease' in their children, as on the EQ-5D parents reported significantly lower health states ($P=0.02$) and quality of life ($P=0.02$) than ventilator-dependent children. Although in contrast, no significant differences were reported by parents in the visual analogue rating of their children's health and parents' values were not significantly different to those recorded by a general population of school children.

One unexpected finding was that two thirds of ventilator-dependent children and parents made a differentiation between health and quality of life. Amongst children, one third thought their quality of life was better than their health and one third thought their health was better than their quality of life. This differentiation is not alluded to in the quantitative literature and warrants further investigation.

Eiser and Morse (2001), in their systematic review of quality of life measures in chronic diseases of childhood, looked at the extent to which child self-reports corresponded with assessments made by their parents. They highlighted a number of methodological issues concerning the development of new measures of quality of life. They suggested that it was standard practice to determine the concordance between child and proxy ratings and if concordance was poor then the measure was viewed as inadequate. There are clearly problems with this argument as experience dictates that children and parents do not agree about many issues in life. Eiser and Morse (2001) found some evidence for greater concordance between child and parent ratings for observable behaviours such as physical functioning, and less for non-observable such as emotional functioning and social quality of life. In the current study this was not borne out entirely, as there were significant group differences between parents and children on the KINDL domains of Self-esteem (positive), Emotional-wellbeing (negative) and Disease (negative), and Health State (Physical functioning) on the EQ-5D (negative). These findings do however add weight to the argument that parents and children make different judgments concerning the child's health and quality of life that may not necessarily indicate that the measures are inadequate.

Although numbers were low, there did appear to be a greater correlation between KINDL scores of 13-19 year olds and parents than 4-12 year olds and parents. In contrast, with the EQ-5D, when asked to identify whether a child needed help to care for themselves, almost all parents and children differed in their assessment. The use and validity of proxy ratings is a very under-researched area, especially for children with such complex needs and impairments. More research with greater numbers of children needs to be undertaken to look at whether concordance is greater on some domains than others; whether parents rate their child's quality of life to be better or poorer than their child's own ratings and to what extent the degree of concordance is affected by the child's age, gender and illness status.

The third hypothesis was that ventilator-dependent children living in hospital would report significantly lower health-related quality of life than ventilator-dependent children living at home. Numbers were low so results need to be interpreted with caution. Findings from the limited data available indicate that children at home have higher levels of health-related quality of life on the EQ-5D and KINDL than children in hospital.

Strengths and weaknesses of the study

This study was obviously limited by low numbers, and the advisory group were correct in their assumption that more meaningful data would have been obtained if ethical concerns could have been overcome to allow a survey of the entire known population. This study was also undertaken with children and young people at different stages in their life course and apart from the fact that the children all used a ventilator there were variations concerning diagnoses, level of dependency on ventilation and life expectancy. For example, some children had been relatively recently placed on long-term ventilation to palliate symptoms of respiratory distress towards the end of their lives whereas others had been on ventilation since infancy, knew of nothing else and were relatively well. Heterogeneity in a small sample may introduce an unacceptable level of bias and a larger sample is required to look at the impact of different variables on outcome.

With a heterogeneous and small population of children such as this one, it is very unlikely that a group-specific instrument will be developed. However, Eiser and Morse (2001) suggest that generic measures are preferable to disease-specific measures where decisions are made involving allocation of resources in a public health context. The sample included more boys than girls and this reflects the fact that males are more likely to suffer from accidents during childhood and during the teenage years gender specific diseases such as muscular dystrophy that only affect boys come into play (Perrin, 2002). In the national database there are also more boys than girls, so the sample reported here reflects the wider population (Jardine *et al.* 1999).

There were also a number of problems encountered with the KINDL instrument that cast some doubt on the rigour of the development and translation of the instrument. As previously described, the German translation proved problematic with small sections of the instrument having to be modified, and the computer generated version could not be used at all with children as corrections to the software could not be made. There are many instances in the literature describing problems with the translation of instruments into different languages (for example, Sperber, 2004). To overcome problems with translation, it is recommended that instruments are translated into the 2nd language by native speakers and then translated back into the 1st language for comparison to ensure the meaning and context are exactly the same. With the KINDL this clearly has not happened and the instrument requires attention to correct these small but significant mistakes.

The KINDL team kindly provided a software programme in English to analyse data. Unfortunately, the Syntax files contained variable labels in English with more characters than SPSS could handle and problems were encountered in re-naming variables in preparation for uploading into SPSS as some of variables were assigned the same label. The KINDL team, however were helpful in correcting these issues and provided a second software programme to analyse data. These problems were unexpected as the KINDL has supposedly been exposed to all the checks and balances required of instruments that have been translated into different languages. Nonetheless,

these unexpected problems are worrying and cast doubt as to the overall rigour and conduct of the translation into English.

Although the KINDL was designed for children from age 4 years old, in practice, most children under 6 could not cope with the questions, even though a smaller and simpler version has been designed specifically for this group of children. The instrument had an appropriate reading score for children of this age (Flesch, 1918; Weeks and Wallace, 2002), and there was provision for the items to be read to children. It may be that ventilator-dependent children who are known to have missed out on a lot of schooling are not as advanced as the general population of school children, coupled with the fact that some ventilator-dependent children have varying degrees of learning difficulty (Noyes, 1999a).

Despite the methodological weaknesses, this study provides new quantitative data on the health-related quality of life in ventilator-dependent children. A strength of the study is that both the KINDL and EQ-5D have been tested with substantial numbers of school children. The importance of having normative data was demonstrated by the apparently high incidence of pain and depression in the general population of school children. For example, with the EQ-5D, over 28% of school children reported they were worried, sad or unhappy: the percentage of children reporting problems also increased significantly with age. In the review of the literature, Lumberg *et al.* (2001) used their own questionnaire that had not been tested with a general population of children and reported that older children were significantly less emotionally content than younger children, which they attributed to differences in reaction to ventilator-dependency. If Lumberg *et al.* had access to normative data they may not have come to this conclusion.

One of the major strengths of this study was that children and young people were enabled to complete their own questionnaires regarding their perceptions of their own health-related quality of life. Two instruments were included in this study and the burden appeared acceptable to children aged 6 and over; although for children who did not use speech as a method of communication it took far longer to administer than indicated in the user manuals. It would not

have been possible to include any more instruments in a battery to measure related concepts as the burden to children would have become unreasonable.

The data collection was carried out very carefully with children and young people who might otherwise not be included in research because of their perceived impairments, especially in relation to communication. The same researcher administered the questionnaires to children in a consistent manner and ensured that parent(s) did not influence their children's responses. This is a model that could be replicated in future studies and reinforces that fact that, when enabled to do so by a specially trained researcher, it was feasible to include children with high levels of impairment in this type of research.

The suitability of using instruments that are not designed specifically for use with disabled children is an important methodological consideration. A discussion of these issues will be presented following the synthesis of qualitative and quantitative data around health-related quality of life of ventilator-dependent children in Chapter 4.

Synthesised findings derived from the 4 research themes will be reviewed in an evidence based and policy context in Chapter 9 and further research directions will be outlined in Chapter 10.

3.6 Summary

Chapter 3 focused on measuring the health-related quality of life of children dependent on ventilators (theme 2). Issues around quality of life were explored through test cases where treatment had been withheld or withdrawn from people dependent on ventilators. Although issues around quality of life were prominent in the legal arguments, judgement in each case was made on points of law and the assessment of quality of life did not feature in the deliberation. The few studies that have attempted to measure the health-related quality of life in ventilator-dependent children were reviewed; these were found to be methodologically flawed and of poor quality.

The chapter then reported a quantitative study underpinned by the medical model of disability measuring health-related quality of life in 35 ventilator-dependent children and young people. Two generic tools were used and when appropriate both child and proxy estimates were obtained. Findings revealed that ventilator-dependent children had significantly lower health-related quality of life than school children; their self-reports varied from those of their parents, and children living in hospital had a lower health-related quality of life than children living at home. These data provide new evidence concerning the health-related quality of life of ventilator-dependent children. The chapter concluded by comparing findings with published literature and discussing the strengths and weaknesses of the study.

The next chapter continues to focus on health-related quality of life in ventilator-dependent children (theme 2) with qualitative methods.

Chapter 4

Describing the health-related quality of life of children dependent on ventilators using qualitative methods: A life worth living?

4.0 Introduction

Chapter 4 is the second chapter in the sequence focusing on the health-related quality of life of children dependent on ventilators (theme 2). In this chapter, qualitative methods that are underpinned by the social model of disability are put forward as an underused approach in health services research to define and explore health-related quality of life. The few qualitative studies that have been undertaken with children dependent on ventilators are then reviewed.

The chapter goes on to report a study that aimed to define and describe the health-related quality of life of children dependent on ventilators from the perspective of the children themselves. Proxy accounts of children's health and quality of life are also explored with parents. In addition, findings derived from exploratory fieldwork with professionals are included to supplement the views of parents and children.

4.1 Exploring health-related quality of life with qualitative methods

There are many advantages offered by qualitative methods when exploring outcomes with users. Qualitative methods were identified during exploratory fieldwork as the preferred approach by ventilator-dependent children and their parents. Qualitative research is a form of social inquiry that focuses on the way people interpret and make sense of their experiences and the world in which they live. Qualitative methods take a more holistic approach than quantitative measures by exploring the quality of life experience and reciprocal interactions

between people and their environments as perceived by the interviewee, as opposed to focusing entirely on their medical conditions and impairments (Alexander and Willems, 1981). The latter are used only to help account for differences in the quality of experience that interviewees report.

Hill and Harris (1993) suggest that qualitative methods may be particularly relevant to researchers working in a relatively under-researched environment, where a package of care rather than a discrete intervention is offered, and in situations where the patients have complex and multiple health problems. This is particularly the case for children who are dependent on ventilators, where the intervention and care package are complex and outcomes for this group have yet to be researched in any depth.

The author has also co-founded the Cochrane Qualitative Research Methods group and has a particular interest in using qualitative methods to help explain and interpret quantitative outcomes. Given the emerging status of this new population of children, it seemed appropriate to use qualitative methods to identify indicators of outcome that need to be monitored on a regular basis and to ensure that the priorities of the children and their families were being addressed. In-depth interviews with children and their families may also assist with the interpretation of quantitative health-related quality of life data which is difficult to unpack or assist in the interpretation of meanings that children and their families attach to particular items (Morse and Field, 1996).

It was therefore decided to undertake a qualitative study to complement the quantitative measurement of health-related quality of life in ventilator-dependent children described in Chapter 2, and then undertake methodological work to synthesise both quantitative and qualitative outcomes around the health-related quality of life in this group of children (which is described in Chapter 5).

4.2 What is already known about health-related quality of life and children dependent on ventilators? A summary of the qualitative literature.

A systematic search of the qualitative literature was conducted in order to shape the qualitative study reported in this chapter. Prior to the commencement of the current study no studies were identified; in fact very little research of this nature has been carried out with children generally.

One small study (Baumgardner and Burta, 1998) was identified subsequently, that explored the quality of life of children dependent on ventilators with parents, employed carers and siblings. This study did not elicit the views of children who were themselves dependent on ventilators. This dearth of qualitative literature probably reflects the emerging status of this population and that much of the research to address important questions has yet to be undertaken.

Baumgardner and Burta (1998) explored quality of life using focus groups with 12 parents, 12 employed carers and 6 siblings of children dependent on ventilators in Wisconsin.

They reported that quality of life was seen as a relative term, defined within the family, dependent upon the availability of services, and expressed in terms of physical comfort, function and integration of the child into the family, and community. Home nursing was highly valued despite lack of privacy. Cuts in services prompted siblings to fear poverty and the divorce of their parents.

Three themes with an ethical dimension emerged from the parents and healthcare workers group. Around the theme of communication, parents said that during their child's acute medical care, they received inadequate, ineffective or overly optimistic views of their child's future (particularly from doctors). Parents said that they received insufficient early contact with parents of a similar child. Healthcare workers said that they believed parents may initially make overly optimistic interpretations of initial information, or be inadequately prepared to receive certain information. Quality of life emerged as

separate theme, especially from the perspective of parents. Parents said that discussions around quality of life were not included or addressed in discussions about their child and that a long-term picture was frequently avoided, indeed parents themselves seemed to be avoided by physicians once acute medical treatment had nothing more to offer. A focus was placed on survival rather than quality of life and the effects on the rest of the family was inadequately or inconsistently addressed. The third theme was around the ethics of decision-making to initiate long-term ventilation. Parents said that their religious beliefs were frequently ignored, with doctors focusing on 'hope' and talking about survival as an outcome, as opposed to quality of life. Parents felt that they were not empowered to make decisions to discontinue life support, and some parents viewed long-term ventilation as experimentation. In contrast to parents who appeared to want some level of input into decision-making concerning their child, healthcare workers said that parents should never have to make such decisions about their child. It was suggested that fear of litigation may drive treatment decisions made by doctors.

This study meets some but not all quality criteria concerning the conduct of qualitative research as outlined in the Critical Appraisal Skills Programme (CASP) assessment framework. (CASP, 2002). The major weakness is the lack of consultation with children and young people who use ventilators, and the small sample size that limits the generalisability of findings. The lack of consultation with children has resulted in findings being skewed towards the impact of the child on the family as opposed to a proxy exploration of the quality of life of the child.

Studies undertaken with young adults

Given that no studies had been undertaken with ventilator-dependent children, the literature search was expanded to look at studies that included young adults in the sample, as the proposed empirical study would include young people under 20 years of age. The needs and experiences of young people going through transition to adulthood would be different to those of younger children and therefore the exploration of this literature was justified on methodological

grounds. Studies undertaken with a middle or old age adult population were excluded as methodologically it is not known if findings are relevant to children and young people.

No studies were identified prior to the current study, but after completion of data collection the following study was published on the internet. In 2002, the International Ventilator Users Network (IVUN) published a qualitative study that included interviews with 26 Canadian adult users, 5 of whom were age 21-25 and had been dependent on ventilators since childhood (Brooks *et al.* 2002). The study examined the concept of health-related quality of life from the perspective of users of mechanical ventilation and described how these perceptions evolved over time.

Brooks *et al.* found that users felt that mechanical ventilation was associated with stigma, especially around disability and what constituted 'normal' healthy lifestyles. Users thought that medical practitioners, and the general public tended to regard the use of ventilation as an intrusive burden, a form of life support that technologised the body to compensate for loss of function. In contrast to the parents in Baumgardner and Burta's study (1998), they did not say that their parents or spouses regarded the use of ventilation as an intrusive burden. Users regarded ventilation as a form of assistive technology, similar to a wheelchair. It was perceived as a benefit to independent living, enhancing energy and overall health. Users considered themselves to be healthy and enjoyed a high quality of life, thus countering societal and some parental assumptions about the use of mechanical ventilation and disability.

Although the findings are useful in providing an insight into a very small number of young adults' perspectives, which indicate some differentiation from the perspectives of healthcare professionals and parents, the study contains some methodological weaknesses concerning the sample. The researchers accept that numbers were small and participants recruited to the study were not representative of the overall population as they had been recruited through advocacy and consumer groups and were connected and involved in society. People were also excluded if they were able to move around independently

without the aid of a wheelchair which meant that the resultant highly dependent sample represented a section of the overall population of ventilator users. Only 5 participants were aged 21-25 and they tended to be successful, active individuals who had adjusted to home ventilation use, accommodated it into their daily routines and moved forward with their lives. They were almost universally positive about using mechanical ventilation and regarded themselves as healthy, active individuals living fulfilling lives.

Although this study was conducted rigorously, the researchers did not seek out deviant cases to explore contradictory positions and the research was funded and carried out by a user organisation committed to advocacy and ventilator users, which introduces the potential for bias in the collection and interpretation of data. This study meets all quality criteria as outlined in the CASP assessment framework apart from the aforementioned aspects of the sample (CASP, 2002).

Quality of life of primary care givers and families

Another group of studies has explored the quality of life of parents and families in general, as opposed to the health-related quality of life of the child *per se*. At the beginning of the empirical work reported here, Sudbury and Noyes (1999) published a comprehensive review of this literature which summarised the considerable negative social, emotional and environmental consequences for the family and primary caregivers of caring for a 'technology-dependent' child at home. The literature was challenging to unpack as many of the samples included children with a range of technologies and disabilities, most of which were less complex than long-term ventilation.

Some of the recurring themes in the wider literature around technology-dependency in children and the impacts experienced by parents were:

- Sleeplessness and the strain of 24 hour vigilance;
- The experience of the healthcare system as a challenge not a support;
- The loss of privacy when nursing staff were providing care in the home;
- Dissatisfaction with lack of space in the family home;

- The loss of independence and varying degrees of social isolation due to the responsibility of caring;
- Feelings of lack of control especially when dealing with employed nurses and services;
- Behavioural and psychological problems in some but not all children and siblings, and
- A higher than average incidence of family breakdown leading to divorce.

For a more in-depth discussion of these issues reference can be made to Sudbury and Noyes (1999).

Factors that influenced the qualitative empirical study

During exploratory fieldwork it became clear that ventilator-dependent children and their parents were keen to be involved in a study that explored their views and experiences around the health and quality of life of the children. Review of the literature found that no qualitative studies specifically around describing the child's health and quality of life had been carried out with children and young people who were dependent on ventilators prior to the commencement of the current study. This lack of evidence around the exploration of health and health-related quality of life of children dependent on ventilators represented a significant gap in the evidence base and warranted further investigation. In consultation with the advisory group set up to guide this part of the research plan, it was decided that it would be a priority to undertake some research with children and their parents and offer some recommendations for improving future qualitative research activities designed to explore health-related quality of life in this group of children. The next section will report the qualitative study undertaken subsequently.

4.3 Qualitative study exploring health-related quality of life with children dependent on ventilators and their families

Selected findings around the health-related quality of life of 53 ventilator-dependent children and their families is reported here.

Method

Aims

The over-arching aims of the qualitative study were to:

- Describe children's views and experiences of their health, social life, housing, education, the services they received, and their overall quality of life;
- Ascertain if children's health, social, housing and educational needs were met, and describe the impact of met and unmet needs on their quality of life, and
- Describe children's aspirations for the future, and ascertain what (if anything) would improve their quality of life.

Research Question

The aspect of the qualitative study reported here was designed to explore the following research question:

How do ventilator-dependent children and their parent(s) view the child's health and quality of life and what impacts do health, social and education services have on their quality of life?

Theoretical perspective

As little (if any) research had been undertaken to describe the sociological construction of ventilator-dependency in childhood, and much of the previous investigation had been within the discourse of biomedical disease, it was decided that qualitative research methods should be employed that allowed for a more progressive construction of the phenomenon of ventilator-dependency in childhood.

It was decided to use three theoretical perspectives to underpin this qualitative study. First, phenomenology using a Heideggerian approach to data collection was considered the most appropriate philosophical approach to describe how

the children interpreted and rationalised the quality of their 'ventilator-dependent' life and their health (Heidegger, 1962). With Heidegger's phenomenological inquiry, the central concern was to establish how ventilator-dependent children self-interpreted their existence in relation to their world. Parents' proxy views of their child's health and quality of life were also ascertained.

Second, a framework for assessing age appropriate 'needs' was adapted to guide the interview process (Pickin and St. Leger, 1993). This lifecycle approach was used to frame the different needs of infants, children and young adults at various key stages throughout childhood. Interviews explored children's met and unmet needs concerning their: health; social life; environment; education, and their aspirations for the future. Finally, data collection and analysis was underpinned by the social model of disability.

Ethics

Ethical issues were as described in Chapter 2.

Sample

Sampling took place in 2 phases: first for the exploratory qualitative study and second for the case studies. Qualitative interviews from the 2 phases were then combined into one sample for the purposes of analysis.

The 2 phases had slightly different sampling strategies:

Phase 1: Exploratory qualitative study

A theoretical sample of 18 children and young people who were aged 6 to 19 years and were dependent on a ventilator were recruited via 9 hospital consultants. This sample excluded children under 6 years as the main purpose was to describe the children's own perceptions and it is more difficult to engage in meaningful conversation with children under 6. To ensure that the sample represented a range of children and their families, it was decided to develop a

theoretical sampling strategy in order to minimally include the following demographic characteristics in the final sample and among those interviewed:

- Three children who had communication difficulties;
- Three children from minority ethnic backgrounds;
- Representation from both boys and girls;
- Representation from children who required assisted ventilation for 24 hours per day and children who used it for less than 24 hours per day;
- A mix of children who lived in hospitals, domestic settings and 'looked after' children in the care of social services, and
- Representation of parents' views, including fathers, as they are under-represented in research studies.

Phase 2: Case studies

It was decided to undertake case studies concerning 35 index children and young people who were ventilator-dependent and aged from 1 year to 19 years. The children and their families were recruited via 11 hospital consultants in the UK. Once again, to ensure that the index cases represented a range of children and their families, it was decided to develop a theoretical sampling strategy in order to minimally include the following demographic characteristics amongst the index cases and those interviewed:

- 10 children age 5 and under;
- 10 children age 6-12 years;
- 10 children and 13-19 years;
- Representation from both boys and girls in each age band;
- Representation from children who required assisted ventilation for 24 hours per day and children who used it for less than 24 hours per day in each age band;
- A mix of children who lived in hospitals, domestic settings and 'looked after' children in the care of social services in each age band, and
- Representation of parent(s)' views.

Recruitment in phase 1 and 2 was successful (Figure 4.1) and all of the theoretical requirements of each sample were satisfied apart from including a 'looked after' child in the aged 5 years and under group. The reasons for this will be explored in the discussion section at the end of the chapter. The resultant combined sample shown in Figure 4.1 included 53 ventilator-dependent children (20 girls and 33 boys); 50 mothers and 17 fathers. Nine children lived primarily in hospital and the remainder lived in domestic settings. Twelve families were from ethnic minorities and 3 children were 'looked after' by social services. Six children needed a carer to help them communicate, 1 young person had no speech or established method of communication (parent read upper lip movement) and 22 others had some difficulties with communicating, such as having speech that was affected by the ventilator and/or their tracheostomy. For 12 families, English was not their first language: in almost all cases an interpreter was not required as their understanding of spoken English was reasonable or good.

Data collection

Fifty-one face-to-face focused interviews were conducted with 35 young people aged 5-19 years where they were living; some children with profound communication impairments were interviewed on 2 occasions. Seven children used talk and draw/play techniques to communicate their views and experiences (Mahon *et al.* 1996). It was not possible to engage with 5 children and young people aged 6-19 years as they had profound sensory impairments and the author did not have the relevant skills to interpret their ways of communicating. Of this group, it was possible to observe briefly 2 children where they were living and proxy reports were obtained from parent(s). Nine children were too young to be interviewed but it was possible to observe them where they were living. The remaining 4 were at school or unavailable when interviewing took place: as before, proxy reports were obtained from their parent(s).

Interviews were conducted with 50 mothers and 17 fathers of 50 children and young people. The remaining 3 children were jointly in the care of social

services and had some contact with their families but they were not available when interviewing took place.

Broad, age appropriate open-ended questions were used to elicit information with probing for further detail. A topic guide provided general direction for the interview and children were encouraged to direct the conversation into areas of importance for them.

Whenever possible interviews were tape recorded and transcribed. A field diary was used to record extensive notes during the study.

Figure 4.1. The sociodemographic characteristic of the sample.

Age range	No	Sex	Diagnosis	Type of ventilation	Location of child	Lone parent	Ethnic minority	Looked after	Child interview	Parent interview
5 years and under	1	F	Spinal injury	PPV tracheostomy (24 hrs)	hospital	✓	✓			✓
	2	M	Congenital	PPV tracheostomy (24 hrs)	home					✓
	3	M	Congenital	PPV (when asleep)	home	✓	✓			✓
	4	F	Congenital	PPV (when asleep)	home	✓	✓			✓
	5	M	Congenital	PPV tracheostomy (20 hrs day)	hospital					✓
	6	F	Congenital	PPV mask (night time)	home				✓	✓
	7	M	Congenital	PPV tracheostomy (when asleep)	home	✓				✓
	8	F	Congenital	PPV tracheostomy (when asleep)	home		✓			✓
	9	M	Congenital	PPV tracheostomy (24 hrs)	home					✓
	10	M	Spinal/head injury	PPV tracheostomy/PPD (24 hrs)	hospital				✓	✓
	11	M	Congenital	PPV tracheostomy (20 hrs day)	hospital		✓			✓
6 – 12 yrs	12	M	Congenital	PPV mask (when asleep)	home				✓	✓
	13	M	Congenital	PPV tracheostomy (24 hrs)	home					✓
	14	M	Spinal injury	PPV tracheostomy (24 hrs)	hospital			✓	✓	
	15	F	Congenital	PPV mask (when asleep)	home				✓	✓
	16	F	Congenital	PPV tracheostomy (24 hrs)	hospital	✓			✓	✓
	17	F	Congenital	NPV perspex chamber (when asleep)	home				✓	✓
	18	M	Spinal/head injury	PPV tracheostomy (24 hrs)	home				✓	✓
	19	M	Congenital	PPV mask (when asleep)	home				✓	✓
	20	M	Congenital	PPV tracheostomy (when asleep)	home	✓				✓
	21	F	Spinal/head injury	PPV mask (when asleep)	home				✓	✓
	22	M	Spinal/head injury	PPV tracheostomy/PPD (24 hrs)	home		✓		✓	✓
	23	F	Spinal/head injury	PPV tracheostomy (24 hrs)	hospital				✓	✓
	24	M	Congenital	PPV mask (night time)	home	✓				✓
	25	F	Congenital	PPV tracheostomy (night time)	home		✓		✓	✓
	26	M	Congenital	PPV mask (Night time)	home	✓			✓	✓
	27	M	Spinal injury	PPV tracheostomy / PPD(24 hrs)	home				✓	✓
	28	M	Congenital	PPV mask (when asleep)	home				✓	✓
	29	M	Spinal injury	PPV tracheostomy (24 hrs)	hospital	✓			✓	✓
	30	F	Congenital	PPV mask (when asleep)	home				✓	✓
	31	F	Spinal injury	PPV tracheostomy (18-24 hrs)	home				✓	✓
32	M	Congenital	PPV tracheostomy (24 hrs)	home		✓		✓	✓	
33	F	Congenital	PPV tracheostomy (when asleep)	home				✓	✓	
34	M	Congenital	PPV mask (when asleep)	home					✓	
35	M	Congenital	PPV tracheostomy (24 hours)	home		✓		✓	✓	
13 – 19 yrs	36	F	Congenital	PPV mask (when asleep)	home	✓	✓		✓	✓
	37	M	Spinal injury	PPV tracheostomy / PPD (24 hrs)	home		✓		✓	✓
	38	F	Congenital	PPV mask (when asleep)	home	✓				✓
	39	M	Spinal/head injury	PPV tracheostomy (24 hrs)	home		✓		✓	✓
	40	F	Congenital	PPV tracheostomy (when asleep)	home	✓			✓	✓
	41	M	Spinal injury	PPV tracheostomy/ PPD (24 hrs)	home			✓	✓	
	42	M	Congenital	PPV mask (night time)	home				✓	✓
	43	M	Spinal/head injury	PPV tracheostomy (24 hrs)	hospital				✓	✓
	44	M	Spinal injury	PPV tracheostomy/ PPD (24 hours)	home				✓	✓
	45	M	Spinal injury	PPV tracheostomy (24 hrs)	home			✓	✓	
	46	M	Spinal injury	PPV tracheostomy (24 hrs)	home				✓	✓
	47	F	Congenital	PPV mask (night time)	home				✓	✓
	48	F	Congenital	PPV mask (night time)	home					✓
	49	M	Congenital	PPV nasal pillows (night time)	home	✓			✓	✓
	50	F	Congenital	PPV mask (night time)	home				✓	✓
	51	M	Congenital	PPV mask (night time)	home				✓	✓
	52	F	Spinal/head injury	PPV mask (night time)	home				✓	✓
	53	M	Congenital	PPV mask (night time)	home				✓	✓

Key: PPV: positive pressure ventilation PPD: Phrenic pacing device

Data analysis

The majority of the interviews were recorded on audiotape except where permission was refused (N=3); in the latter cases notes were taken. Interviews were transcribed in full (where recorded). Transcripts and notes of interviews were anonymised and entered on to computer for analysis using Atlas Ti™ version 4.2 (Atlas Ti, 2002). Where appropriate, these data were analysed in conjunction with observations recorded during fieldwork and the children's drawings. It was decided to use principles and procedures of the 'framework approach' to guide data analysis as outlined by Ritchie and Spencer (1995). The framework approach is particularly suitable when conducting applied policy research whereby the aims and objectives have been shaped by specific information requirements (in this case the need to describe how children conceptualised their own health, quality of life, and met and unmet needs). The five key stages to this method of thematic data analysis are:

- Familiarisation;
- Identifying a thematic framework;
- Indexing;
- Charting, and
- Mapping and interpretation.

During the last two stages of data analysis when all data have been sifted and charted according to core themes, it is then possible to pull together key characteristics of the data, and to map and interpret the dataset as a whole. Concepts, constructs and explanations around the children's health, their quality of life, and their needs were defined, described and mapped against the aims and research question.

As several of the 18 children in the initial exploratory qualitative study did not use speech as a method of communication, the Joseph Rowntree Foundation provided extra funds to employ a graphic artist to depict in cartoon format some common views and experiences. These cartoons were returned to interviewees for feedback to see if they accurately represented their views and experiences and they were evaluated highly. Cartoons also served to convey the sarcastic

wit and humour demonstrated by young interviewees. There is only sufficient space to present 5 cartoons here. For a report containing all of the cartoons developed with children, please refer to Noyes (1999a).

Exploratory fieldwork with professionals

In addition, findings derived from exploratory fieldwork with professionals were included to supplement the views of parents and children. The methods concerning exploratory fieldwork were described previously in Chapter 2.

Findings

It was clear from visiting the children and young people and observing them where they were living that they had a far from 'ordinary' childhood. Their life experiences were shaped by periods of illness and wellness, disability, strict routines around medical device use and other interventions, adult surveillance, constant risk management concerning activities and frequent contact with hospitals and various professionals.

Data reported here are limited to key and sub-themes around the children's and their parents' constructs around the children's health-related quality of life.

Key themes around children's health-related quality of life

Children's constructs around health

Feeling better on the ventilator

Irrespective of the underlying diagnosis, reason for ventilation or life expectancy, children's perceptions around health and ventilator use had one common feature – everyone who was able to express a view said that their ventilator made them feel better and if they had sufficient 'breath' then they were able to do other things that they enjoyed.

Level of dependency on the ventilator

The children and young people had varying levels of dependency on the ventilator. This ranged from children and young people that were dependent 24

hours a day, to those who only used ventilation at night or whenever asleep. Ventilator use could also increase during periods of acute illness when children and young people suffered greater difficulty with breathing.

The level of ventilator-dependency appeared to have no bearing on the children's perceptions regarding their overall health, although it did have an impact on their quality of life experiences. Children and young people said that things were less complicated if they were not totally dependent on the ventilator and could manage without it for periods of time during the day. This allowed greater freedom to participate in activities with less need for adult surveillance, which had a positive impact on their quality of life.

Staying healthy

Once discharged home from hospital after the initiation of ventilation, most of the young people seemed to be relatively well and enjoyed what they considered to be *'good or fairly good health'*. Most children suffered from infections such as chest and urine infections, and periods of acute illness; many described episodes of having to stay off school or being readmitted to hospital. Seven children however had not returned to hospital in the past year apart from regular routine check ups. Most did everything in their power to avoid hospitals and the thought of returning even for a check up was considered one trip too many.

Parents' constructs concerning their children's health

Parents' views generally mirrored those of their children although they were more likely to use words such as *'mixed'*, *'up and down'* and *'mainly good'* to describe their children's health. It was difficult to assess the objectivity of 2 mothers who had campaigned for their children to be put on ventilators and described their children's health and quality of life on ventilators as *'perfect'* despite the fact that their children had life-limiting conditions, serious health problems and multiple impairments.

It was also difficult to ask some of the questions around quality of life and decision-making when the parent and child were sitting together as the parent

was unable to extricate him or herself from their caring role during the interview. It was not possible in most interviews to explore whether parents were involved in discussions with doctors concerning the decision to initiate or continue with long-term ventilation. One parent who was interviewed on their own said that they questioned whether their child should have been treated with long-term ventilation.

Perceptions of doctors

Doctors reported that little was known about the long-term outcomes or health-related quality of life of these children who survived with varying degrees of physical and intellectual impairments and a requirement for life-long expensive packages of care. Doctors frequently acknowledged that they had faced impossible dilemmas with little or no evidence to guide their decision-making.

Staff working in paediatric intensive care units spoke of their feelings of frustration when a child remained dependent on ventilation. They had to deal with blocked beds and turn away critically sick children from their units who may have made a full recovery had they been given access to intensive care. Few doctors were said to be keen on managing children dependent on ventilators as they preferred to work with critically sick children who could benefit from their intensive care skills.

Doctors said that they faced difficult moral and ethical dilemmas in terms of what to do in the best interests of the child (the central tenet of all child care law). From a human rights perspective, there was also said to be a polarisation of argument between individuals and organisations who were campaigning for every child whatever their impairments to have access to long-term ventilation, versus the perspective of some clinicians who held differing opinions concerning their interpretation of what constituted 'the best interests of the child' in terms of quality of life and associated burden on the family and state.

Conversations concerning the test case to withdraw treatment from a child with a severe life-limiting genetic disease (Chapter 3) revealed that this was an extremely stressful legal process for all concerned; not all doctors said they

would be prepared to go to Court in similar situations. During the course of this work, the author met some children with a similar condition to child C who had been offered treatment despite the outcome of the test case.

The view expressed by one doctor *'I don't know whether I'm prolonging life or prolonging death in these difficult cases'* was typical of the views expressed by doctors generally who felt that they had been pressurised (mostly by parents) into offering long-term ventilation to children with multiple impairments and a poor prognosis. Doctors talked of individual cases of children that they had come across during their careers whereby long-term ventilation had been used inappropriately in their view. For example, one doctor described the case of a child who was in a persistent vegetative state and had been treated with long-term ventilation in an acute hospital for the past decade.

Other professionals viewed long-term ventilation as an intervention that conveyed considerable health benefits for most of the children and young people they had managed. These professionals had gained considerable experience with this new emerging population and had seen children flourish at home with their families.

Constructs around the impact of morbidity, co-morbidity and length of ventilator-dependency in relation to their health and quality of life

Children's responses depended on the reason for their long-term ventilation and how long they had been ventilated.

Ventilated since infancy

Children who had been dependent on ventilation for all or most of their lives and who knew of nothing different appeared to have adapted to their circumstances. These children and young people talked about the barriers that society put in their way as affecting their quality of life as opposed to their physical and learning impairments and dependency on ventilation *per se*.

Ventilated following a serious illness or accident

Children who had previously been well, but had become dependent on ventilation following a serious illness or an accident and could remember themselves in their pre-illness state frequently talked about wanting to regain a physical function that they had prior to the accident such as wanting to walk again or get off the ventilator. They too were angry about the societal barriers that were put in their way to prevent them from participating fully in society. Four young people who had experienced serious accidents in the recent past appeared depressed at their current circumstances and their mood had a significant impact on their mental health and quality of life at that time.

Ventilated because of morbid obesity

One child who required assisted ventilation because of morbid obesity talked about their weight and the associated bullying by other children (as opposed to the ventilator which made them feel infinitely better) as the major factor that compromised their health and quality of life.

Ventilation as palliative care

Children and young people with life-limiting conditions who required ventilation towards the end of life to alleviate increasing difficulty with breathing talked about having more energy and generally feeling better because of the ventilator. This group included teenage boys with muscular dystrophy who had been offered assisted ventilation as their progressive muscle wasting made it increasingly difficult for them to breathe. The ventilator had clearly extended their lives but it was ethically difficult and somewhat insensitive to explore quality of life with these children who were all essentially receiving palliative care at the time. Interviews with these children and young people explored the hobbies they enjoyed and things they liked to do. It was clear that being able to breathe more efficiently was considered a good thing and had a positive impact on the activities that children engaged in. None of the children or young people talked about the inevitable point at which assisted ventilation would be ineffective. All their conversations were focused in the here and now and not too distant future.

Children with profound impairments

Parent(s) provided proxy accounts for children with profound impairments, babies and toddlers. Four children and young people had such profound sensory impairments that it was not possible to engage with them (although every effort was made to include as many children as possible). It was sometimes possible to observe these children and young people where they were living. All those observed appeared to demonstrate facial expressions indicating that they were contented and the children responded positively to their parents and carers. Parents were able to interpret their children's facial expressions, behaviours, signs and noises.

Parents of younger children with profound sensory and physical impairments usually described their children's health as mixed and they were all very positive in their proxy accounts about their quality of life. In contrast, parents of older children talked positively about the relative state of their child's health and quality of life but they appeared less certain about the future. Many of these children had life-limiting conditions and ventilation had been added to relieve respiratory distress. One mother said of her teenage daughter who had profound impairments:

'We will listen out for the doctors to let us know when enough is enough, but we are not there yet'

Babies and toddlers

Nine were babies or toddlers and it was only possible to observe them, and ask their parent(s) to provide proxy accounts. All young children who were observed appeared to demonstrate a range of facial expressions and behaviours to communicate their needs; some children made noises, signs and used speech that parents were able to interpret. When settled, the young children looked contented and responded positively to their parents and carers. Parents consistently talked about their children as *'being much loved'* and being cared for by people who provided them with a good quality of life and varied life experiences. Most parents did not compare their child with other non-

disabled children so health and quality of life was seen as a relative thing and unique to each child.

Children's constructs concerning their quality of life

Quality of life was clearly a term that was not included in the vocabulary of most children under 12 years of age. A response during an interview that was typical of many children was given by Navin (aged 12 and dependent on ventilation since an accident age 8):

Interviewer: How would you describe your quality of life?

Navin: 'Mum, what's quality of life mean?'

A reflexive approach was therefore taken to interviewing younger children to explore their quality of life by asking about their life experiences concerning their health, likes and dislikes and the activities children wanted to do, followed by factors that prevented them from engaging in such activities and suggestions as to what would make things better for them.

Quality of the lived experience

The quality of life experiences of children and young people emerged as having a profound impact on their quality of life. Twelve sub-themes emerged:

- Lack of respect and understanding;
- Inability to communicate effectively;
- Inequity of access to health services;
- Prolonged and unnecessary periods of hospitalisation;
- Being enabled to live at home;
- New technologies;
- Availability of accessible good quality and well-coordinated services;
- Availability of accessible and affordable transport;
- Friends and hobbies;
- Access to school and a good education;
- Aspirations for the future, and

- The consequences of being a 'looked after' child.

Each sub-theme will be explored in the subsequent paragraphs.

Quality of lived experience, sub-theme 1: Lack of respect and understanding

Most of the children and young people in this study did not consider themselves as being 'disabled'. They said that they were more disabled by the barriers that people placed in the way of them getting on with their lives. Only the young people who had recently experienced a sudden change in circumstances following an illness or accident said that they were now 'disabled'.

The negative way in which the young people were viewed by nurses, doctors and people generally was highlighted by several of the older children during the interviews. One adolescent summed up typical sentiments:

'You can't have one (a physical impairment) without the other (learning difficulties).'

He felt that he was consistently treated as if he had learning difficulties because he had a spinal injury. Another adolescent girl who had learning difficulties was not happy with the way she was generally treated. She said:

'I look after myself really, I'm not that stupid you know, I'm coming along.'

This perceived lack of respect appeared to have a considerable impact on the self-esteem of the young people who said they did not feel valued.

Quality of lived experience, sub-theme 2: Inability to communicate effectively

For 28 children, the issue that had the most impact on their quality of life was not being able to communicate effectively. Five of the young people did not have an established method of communicating. For some, this was because their speech had been affected following an accident and they had not been provided with an electronic voice synthesiser or augmentative method of communication. Other children and young people had a tracheostomy and this

made speaking more difficult. Not being able to speak clearly was very frustrating and hampered expression of opinions and social interaction. There was a great deal of frustration at the way the young people were treated, especially as nurses and doctors had not been trained to communicate with children who did not use speech, and many professionals did not ask the children for their opinions. One 16 year old boy complained about the way he had been treated:

'What kind of human treats other humans like that?'

The issue of young people having the right to express their opinions was considered very important. Parents acknowledged that a young person's opinions might be different to that of parents and professionals. Not being able to communicate effectively also had an impact on the children's self-esteem and emotional wellbeing as they were prevented from interacting with others and did not feel valued.

Quality of lived experience, sub-theme 3: Inequity of access to health services

All children and young people with the greatest dependency on ventilation and the most complex healthcare needs spent prolonged periods of time in hospital after the initiation of long-term ventilation. These young people described being moved between up to 4 different hospitals, some of which were hundreds of miles from the family home. Most parents said that there was inequity of access to services because of their child's dependency on a ventilator. David's case outlined in Figure 4.2 illustrates a typical pattern of moving around different hospitals and regions that had a major impact on his quality of life and the quality of life of his family.

Figure 4.2. David's case history.

David was born with a breathing problem at his local hospital. Soon after his birth he was transferred to a local neonatal intensive care unit. A few weeks later David was transferred to another specialist neonatal intensive care unit over 100 miles away from his family home. He was then transferred to the children's ward in the same hospital. After several months he was transferred back to a regional hospital nearer to his home. He was eventually discharged home for the first time at 17 months old.

In contrast, children and young people who used ventilation to enhance or boost their breathing with mask ventilation at night and were not reliant on it said that they often only went into hospital for a night or two to adapt to the equipment. Their experiences of the health service were generally more positive as they did not spend prolonged periods of time there.

Lack of rehabilitation services in hospital

Children with the most complex needs said that not all hospitals provided a programme of rehabilitation following the initiation of long-term ventilation. Over half the parents complained that their children were continually treated as a medical case and said that nurses and doctors only talked about ***'what their child could not do and not what their child could do'***. Parents wanted their children to be regularly reassessed and not be left from week to week with little happening. Parents said that interventions to rehabilitate their children such as intensive physiotherapy and speech therapy had a positive impact on their quality of life, but their children frequently received insufficient input from such therapists.

Lack of statutory education in hospital

The children and young people said they were frequently deprived of an adequate education whilst in hospital. They wanted to be educated and did not want to fall behind their peers as it would affect their life chances. The attitudes of staff also varied widely. Jason's mother explained how differently her teenage son was treated in 2 hospitals:

'I asked one of the nurses why isn't he being educated, where is the hospital school service?'

She was taken aside by the consultant and told not to worry about his education. Jason took up the story and related what happened soon after he was transferred to another hospital:

'One of the teachers started with a maths lesson and I said 'I cannot do it', and she said 'why not?' And I said 'well I'm disabled' and she said 'well that's no excuse!'

Many young people expressed that **'education was the only thing that kept them going'** and it instilled some structure into otherwise **'long and monotonous days'**. All of the young people missed significant periods of schooling, in some cases this amounted to a number of years. Hospital teachers were simply not equipped to deliver the national curriculum to young people who spend prolonged periods of time in hospital. Parents said that the quality of the education children received was not monitored and a lack of education had a negative impact on the life chances available to children.

Quality of lived experience, sub-theme 4: Prolonged and unnecessary periods of hospitalisation

Thirty-three of the young people said they had spent between 1 and 6 years in hospital and this was usually one long admission following the initiation of assisted ventilation. All but one of these young people said they spent many months (and some cases years) in hospital when they did not need to be there waiting for their discharge home to be organised. Of the remaining 20 children and young people: 11 had experienced many episodes of being in hospital, and 9 were currently still in hospital awaiting discharge: one of the latter children had been in hospital over 3 years waiting for a foster parent to be arranged.

Sensory overload and deprivation

Children said they experienced both sensory overload and deprivation whilst in hospital and this had a negative impact on their wellbeing. The children and young people did not always have access to play and activities to keep them stimulated and occupied. All of the young people had spent unnecessarily prolonged periods of time in an intensive care unit or high dependency unit as there was no alternative placement. Parents worried about the **'constant horror that goes on in intensive care'**. Eight young people were cared for at some stage in adult wards that did not have suitably qualified children's nurses, toy cupboards, designated play therapists or appropriate soft furnishings. The young people talked about **'a diet of TV and videos'** that was fed to them. It was also noted how late bed time was for very young children in adult wards.

In contrast, being in an adult ward suited the teenagers who did not want to mix with younger children. Mohammed aged 15, said of children's wards:

'They are so noisy, the young ones do your head in.'

Lack of control over children's lives

The young people said that they found it difficult to get out and about and meet friends whilst they spent long periods of time in hospital. A sense of being in limbo and socially excluded was very prominent in many conversations and this had a negative impact on their quality of life. One young person said:

'I am disabled therefore I am not normal, lock me away from the real world so I cannot be a part.'

Sean's story outlined in Figure 4.3 demonstrates one of the more extreme cases whereby he was forced to live away from home for 3 years as the health service was unable to meet his needs.

Figure 4.3. Sean's story.

When Sean was a teenager he was involved in an accident and spent 3 months in his local hospital. It became obvious that he would need long-term ventilation and he was then transferred to a specialist centre over 150 miles away. His mum rented accommodation locally and left the rest of the family to cope at home. Dad commuted to the hospital every weekend. Sean did not have access to a communication system that suited his needs.

His first discharge-planning meeting happened over 1 year later. Sean's health authority were initially unable to fund a care package enabling him to live at home. Sean did not want or need to be in hospital and his parents fought a long battle on his behalf.

Three years after the accident, Sean was discharged home and his family were reunited again. Sean and his family said his prolonged hospitalisation was unnecessary, unacceptable, a waste of public money, a personal financial burden and forced them to live apart for 3 years. Sean also missed out on vital schooling and was deprived of 3 years of his life. Sean is now rebuilding his life at home and has not encountered any major problems since discharge 1 year ago.

As Sean's story illustrates, there were very obvious inequalities in the way hospitals treated these young people. Policies, procedures, checks and routines mediated by nurses and doctors were said by parents (and some of the adolescents) to dominate the children's lives. Some hospitals wanted to maintain control, whereas others empowered the young people and their parents to take more responsibility. One boy described being in hospital for 3 years as an adolescent; once his parents were trained to care for him they went out together to local shops and restaurants. He even described going up in a glider when he was still an in-patient of a high dependency unit.

In contrast another mother was angry that she could not initially leave the intensive care unit (and subsequently the hospital grounds) with her daughter. She said:

'The nurses had to follow you around all of the time.'

If nurses were not available (which was frequently the case) they were unable to go anywhere.

Children who were not enabled to have control over their lives developed a number of psychological and behavioural responses. For example, many parents said that in later life their children remained insecure, mistrusted people and did not like leaving the house. Others described their children as only wanting to associate with adults and avoided contact with other children. Several parents recalled their children would go to any adult, which caused fear for their safety. Some of the children refused to eat food and as a result underwent additional surgical procedures (such as insertion of a gastrostomy tube) to ensure their nutritional intake was adequate.

Parents recounted how some children had developed destructive behaviour and were violent:

'Neither of us can control him, he won't do as he is told, he seeks attention, has temper tantrums, he will throw a chair across the room.'

Parents generally felt that their children's psychological and behavioural problems resulted from spending prolonged periods of time in hospital (principally in an intensive care environment) and having little control over their lives.

All of the young people remembered desperately wanting to go home. They described and gesticulated their boredom, apathy and in some cases their hate of hospitals. One 6 year old said:

'I didn't want to be there any more (hospital).'

Separation from families

The children and young people said that they wanted to see their parents whilst they were in hospital. For parents the most contentious issues were the lack of equity regarding residential accommodation and access to their children. Attitudes varied widely from hospitals that encouraged parents to stay with their child free of charge, to hospitals that did not provide any parental accommodation. Five families talked of having to ***'find and rent accommodation'*** locally in order to be near their children. A frequent complaint from parents whose children were in adult areas of hospital care was:

'They (nurses) would never let you in to see them.'

Psychological impact

The children, young people, and their families, had vivid and lasting memories of hospitals. Jack who was 3 years old at the time of his accident had explicit memories of being in hospital. He said:

'I got run over ... a car got me... and I went to (name of hospital) in a helicopter...and they put that horrible thing in my nose...horrible thing those tubes.'

Almost all of the children in hospital did not have dedicated nurses and were looked after by numerous nurses who came and went with each shift. One mother of a toddler said that a different nurse put her child to bed and another

one got him up for nearly 2 years of his life whilst he was in hospital. Parents heard terms such as '*institutionalised*' used to describe their children.

Losing contact with families

Three children had experienced loss of contact with their families whilst they were in hospital because of complex social issues and this had a very profound negative impact on their quality of life and wellbeing. These young people said they were not always consulted regarding their need for continued contact with their parents and siblings, and nurses did not always actively promote or support their right to live or maintain contact with their family. Those young people who no longer lived with their families said they were uncertain of their life histories and biographies, and one young person did not have these essential memories maintained for him. Some of the young people were losing contact with their language, religion, culture and roots. Those young people who lived in hospital accommodation for long periods of time said that they did not belong to their families any more.

Getting home from hospital

Most of the negative impact of prolonged hospitalisation on the child's quality of life could have been avoided if they had been discharged earlier. Parents said that nurses were not experienced or competent at coordinating their child's discharge. Nurses were said to be frequently hampered by bureaucracy and the failure to agree funding for a package of services to enable the child to live at home. These economic issues will be explored further in Chapters 6 to 8.

Good practice

Several families talked about named specialist hospitals as being 'very good medically' and having 'doctors, nurses, teachers and social workers who knew what they were doing with his condition'.

Quality of lived experience, sub-theme 5: Being enabled to live at home

Despite having to contend with major adjustments, the children and young people said they were very relieved to be home and used words such as

'brilliant', 'excited' and 'could not wait' to describe how they felt about going or returning home.

Home was a strikingly different place to hospital and all the children and young people said that being at home had a positive impact on their quality of life. Sarah (aged 7) who was discharged home for the first time since her birth expressed views typical of many young people. She said:

'I didn't want to be there (hospital) any more. It's nice to be home. It's nice to be with my mum and it's nice to be with my sister.'

Homes were observed to be lively places that were invariably noisy and busy with many people coming and going. Decorations, sounds and smells reflected ethnic, cultural and religious identities. The young people had stamped their own identity on their bedrooms. The impact of the variety of colours, fabrics, textures, furniture, smells and sounds of these rooms provided a stark contrast to a hospital ward. Equipment such as ventilators and suction machines were not obvious.

Efficient and portable equipment

Almost all of the children and young people said that it was important to have modern and portable equipment such as ventilators, suction machines, oxygen monitors and wheelchairs. This was because newer machines tended to be smaller and have a longer battery life, which enabled the children to be more independent and mobile, thereby enhancing their life experiences. The latest machines (especially ventilators) were actually designed for home use and were said to suffer from fewer false alarms, which meant less aggravation.

In an image conscious world, the children and young people wanted the latest designs in wheelchairs, which were not always available through the NHS. A dowdy but sturdy NHS model was simply not cool and lack of choice concerning wheelchairs was a major concern. The children and young people demonstrated that they were discerning consumers; they did not want to be

seen in something that was not 'cool' and they perceived that a 'cool' image was associated with having a good quality of life and 'fitting-in'.

Homes fit for living

Both children and parents said that the design of their home had a major impact on the quality of life of the child. Six families had purpose built accommodation that had been funded from insurance payouts. Three large 'smart' homes in particular were designed by specialist architects and on inspection were superb. The children and young people said that they enjoyed living in these homes as they were enabled by technology and could access all aspects of the accommodation independently.

In contrast, over half the families said their home did not meet their needs. The problems highlighted by the children and young people are summarised in Figure 4.4.

Figure 4.4. Problems encountered with housing.

- Not enough room;
 - Insufficient power sockets;
 - Doorways too narrow for wheelchairs;
 - Lack of space or privacy for rest of family;
 - Lack of space/facilities for carers, and
 - Lack of lifting equipment.
- Ventilator-dependent person was unable to:
- Use toilet or bath;
 - Use wheelchair in house;
 - Access upstairs floors;
 - Access garden, and
 - Control their environment (turn lights on/off etc).

These problems increased as the young person had grown up. Some families were resigned to living with these inconveniences whilst others were looking to extend or move to more appropriate housing. The economic issues related to housing will be explored further in Chapters 5 to 8.

Many children and young people liked visiting the homes of relatives and friends but this presented many difficulties concerning access, which impacted on the child's enjoyment of life. Sunil age 10, who was a wheelchair user, talked about a common problem when he visited his granny: he said that he could not move about in his wheelchair once in the house. As a result visits to his relative's houses had to be short and he did not go as often as he wanted to.

Quality of lived experience, sub-theme 6: New technology

Six children in the current study who required assistance with their breathing 24 hours a day had benefited from a new technology called a phrenic nerve pacing device. This device was implanted in the child's body and stimulated the phrenic nerves to expand and contract the lungs thereby negating the need for the ventilator during periods when the phrenic pacer was turned on. Sam who was 8 years old said that his phrenic nerve pacer was good because he did not have to be attached to his ventilator with bulky tubing that plugged into his tracheostomy; this meant that he had more freedom to move his head and he could wear a greater choice of clothes. His speech was clearer and louder when not having to speak in synchronisation with the ventilator, and he was not so conscious of the distinctive smell that ventilator-dependent children emit from their tracheostomies when open. The phrenic pacer was much quieter than the ventilator and therefore did not distract him or his classmates; as a result he said that his confidence had increased and he felt less stigmatised by the presence of the equipment that maintained his life.

Quality of lived experience, sub-theme 7: Availability of accessible good quality and well-coordinated services

For the majority of children and young people, their quality of life at home was affected by the lack of coordination of care and poor management practices.

The young people described a number of other people who visited them at home (see Figure 4.5). Visits were frequently uncoordinated and the constant interruption and intrusion was a source of irritation. Interviews were constantly interrupted by the phone or door bell ringing. During one 2 hour interview 4 different professionals either telephoned or visited the home unannounced.

Figure 4.5. List of professionals who visited families at home.

- Carers from child's care team
- Respite carers
- NHS Team leader/case manager
- Voluntary sector coordinator
- Social worker
- General Practitioner
- Hospital doctor
- Speech therapist
- Occupational therapist
- Physiotherapist
- Dietician
- District nurse
- Health visitor
- Specialist health visitor
- Psychologist
- Educational psychologist
- Education welfare officer
- Occasionally home teachers
- Home help
- Family support worker
- Solicitor
- Child minder (for siblings)

The impact of employed carers on the child's quality of life

Children and their parents had very different experiences of their employed carers. Some families initially found themselves living with total strangers. Not all families had been able to negotiate appropriate boundaries and significant problems had resulted such as carers taking food from the family kitchen.

Inevitably, a number of problems had been encountered which had a negative impact on the quality of life of the child and their families. These ranged from sickness and absence amongst the carers to inappropriate behaviours. Seven young people talked of these issues sometimes getting *'out of hand,'* or at times being near to *'breaking point'*. They used words such as *'despair,' 'angry,' 'frustrated,'* and *'had enough'* to describe their feelings. The young people talked about the intolerable pressure on them and their parents and how tense the atmosphere at home could be. For example, one teenager recounted how the nursing team leader entered his home:

'She came in and she was having full blown arguments with one of the carers on the stairs. We could hear her in the dining room.'

A number of children and their parents said they had to find ways of coping with additional unnecessary stress as a result of on-going battles with professionals, employed carers and health authorities. They talked about *'having no control'* and *'being depressed'*. Complaining about various agencies featured highly in conversations. Some parents were unable to carry on in full-time employment and the young people talked about their parents need for anti-depressant drugs.

Three quarters of children and young people had designated employed carers to care for them. The young people had much to say about the carers that had been employed to look after them at home. They felt that apart from meeting their health needs carers were very important socially. There was a feeling that young people did not want an entirely professional relationship with carers and many had established strong friendships. The following qualities were considered important in carers, including:

- Similar age (very important they were not too old);
- Good looking;
- Good fun;
- Sense of humour;
- Happy;
- Friendly;
- Kind;
- Likes similar things;
- Specific gender or mix of genders, and
- Ability to listen and learn about their care.

Like all of the young people, Mohammed (aged 15) was very specific about the type of carer he wanted to look after him:

'Nice, not unhappy, not grumpy and happy to look after me.'

Several of the children and young people talked about individual people who had made a big difference to their lives. Amy (age 19 years) said that her team leader was **'absolutely terrific and had become a good friend'**.

In contrast, parents talked about carers who assumed the role of parent, and unwanted intrusion into family activities. One common complaint was that carers ignored parental wishes regarding discipline. One mother recalled:

'We want the nurses to say yes or no and stick to it.'

Risk management policies

Many of the young people and their parents felt that **'unnecessary boundaries were created by professionals'**. Getting the right balance between health and social care did not always happen. Some carers were said to assume that they were in charge in the home and dictated what the young people and their families could and could not do. The young people said that there were far more things carers did not allow them to do. Some bedrooms were set up and run like intensive care units. Brothers and sisters were not allowed in unless accompanied and supervised by a responsible adult.

Some parents and young people highlighted many activities that were censored by overzealous team leaders who felt it was **'too risky'**. One such example was staying in a hotel and visiting a theme park. In contrast other families described the benefits of enjoying a family day out and the excitement of appropriate theme park rides. Lisa (aged 12 years) said she loved going to see a boy band in concert with her family. Her mother encouraged her to experience different things. A number of families would not accept unnecessary limitations on their lives.

The common experiences of children and young people concerning risk management and control are illustrated in cartoon format in Illustration 4.1.

Illustration 4.1. Cartoon indicating that young people said some carers unnecessarily restricted their lives.



Respite care, breaks and holidays

The young people and their parents said that they had different needs regarding respite care, breaks and holidays. For the children and young people, being able to take a break was considered very important and the experience enhanced their quality of life. Most young people had enjoyed going on holiday and appreciated the enormous amount of organising by parents and their employed carers.

Despite visiting some great places, almost all children had experienced a number of problems, including: a lack of options concerning destinations and accommodation; having to transport large amounts of equipment; being unable to get travel insurance; having to pay to take the entire care team on holiday with them; and unexpected equipment malfunction.

Jessica talked about having to cram all her equipment in 2 cars. Many parents recalled that they arrived at the destination, plugged in the ventilator and prayed

it would work and the battery provision would be sufficient if there was a power cut. Some parents were more adventurous and had taken children on long haul flights to more exotic destinations such as Florida. One mother provided a picture taken from a company brochure for a portable ventilator. She described that her experience was nothing like that depicted in the promotional literature showing a child cycling around a campsite towing her ventilator whilst her mother appears to be looking away (Illustration 4.1). Life with a child on a ventilator was far more stressful and she did not dare take her eyes off her child for a second in case she became disconnected from the ventilator. The mother said that her daughter would be unconscious within seconds and dead within 2-3 minutes.

Illustration 4.2. Image from a ventilator manufacturer's promotional material. Original in colour.



In contrast to the children, the biggest issue for many parents was the lack of respite care provision so that they could go away on their own or have an evening off. Taking a break was seen as being absolutely essential and significant problems had been encountered as there was insufficient provision to meet demand and employed care teams could be very inflexible.

Good practice

Eight families said they liked having one key person who did all the coordination between services and organisations (although this did depend on the personality and flexibility of the person who coordinated things). When this

worked effectively, quality of life generally was said to be much better and families experienced less hassle, leaving them more time to enjoy life. Families who had 24 hour medical and mechanical support said they appreciated the peace of mind. Parents and, when appropriate, the young people liked attending and contributing to meetings to review their care package which often took place in their own homes. Teenagers in particular liked being involved in making decisions concerning their care and well managed, personalised and responsive care packages were said to make a big difference to their quality of life.

Quality of lived experience, sub-theme 8: Availability of accessible and affordable transport

Being able to get out and about easily was said to have a positive impact on the child's quality of life. Of the 53 children and young people:

- 19 used a people carrier/van/minibus;
- 20 used a car;
- 13 used black cabs or adapted minicabs;
- 5 used a low level bus;
- 5 relied on relatives;
- 10 relied on hospital transport, and
- 3 walked or sometimes used dial-a-ride.

One young man was learning to drive and hoping to pass his test in the near future.

Over half of the children and young people interviewed said that they had problems getting out and about and this severely limited their enjoyment of life. Some teenagers were not able to use a taxi as there was not enough headroom. Mohammed said that when he was sitting comfortably in his electric wheelchair his head touched the ceiling inside the taxi. Dial-a-ride was singled out as being particularly inflexible. Sarah's mother said:

'We've tried dial-a-ride, but it wasn't successful, they don't go out of the district. I've found them very difficult...They are just not equipped for her.'

Fay and Jack were measured and fitted for a buggy to get around in. These were subsequently found to fail the British safety standard and could not be used in a vehicle. They then had a bureaucratic battle on their hands to get a suitable replacement. Fay's mum said:

'In the end we said we would buy one (buggy), we don't care how much, just get one or we are never going to get out of the door.'

Replacement wheelchairs were singled out as being a major ongoing problem. It was difficult to get appointments to be measured and the NHS supplier took months to deliver the finished product. One young person was waiting for his fifth chair to be delivered in 5 years. His mother said the chairs had not fitted correctly and they had to wait a year from measurement to delivery. Similarly, Jason expressed dissatisfaction about having to wait for so long for his new wheelchair:

'I've been sitting in this crap chair for 4 months, it's doing me no good, I've got spinal curvature now.'

Electric wheelchairs generally had insufficient battery power to meet the needs of young people and several teenagers said that electric wheelchairs supplied through the private sector had far longer battery life to go out and out about independently than those supplied via the NHS.

Quality of lived experience, sub-theme 9: Friends and hobbies

Many children and young people were sad that they were no longer in contact with their friends. Others had difficulty in meeting friends (especially non-disabled friends), of the same age and with similar interests. They felt that this was due to a number of reasons, including:

- Lost contact with friends while in hospital;
- Used to adult company;
- Friends stayed away after young person commenced ventilation;
- Moved house to a different area;
- School was a long way from home;
- Unable to go out alone, and
- Unable to have or go to 'sleepovers' with friends.

Parents tried hard to compensate. For example, Scot's mother had made sure her son participated in a sleepover in the primary school hall. She had to get permission from the health authority, and said it was worth the hassle as Scot really enjoyed it.

A number of young people recalled that things had changed after they became dependent on ventilation. For example, Sunil age 10 said:

'They used to drop by... they used to come here every day... but now they don't... I wanted to be friends... but they didn't.'

Bothers, sisters and carers were a main source of company. Many families said they had become very close and they did a lot of activities together.

In contrast other children and young people said that they had brilliant social lives and went out with their friends all the time. These children and young people appeared to have few problems with transport, lived nearby their friends and spent time off their ventilators during the day.

Quality of lived experience, sub-theme 10: Access to school and a good education

One of the biggest issues that could impact positively or negatively on the child's quality of life and life chances was said to be the child's education. Parents complained that the options open to their children were limited. They said that finding the right school was ***'yet another battle'*** and they had to ***'fight for their children'***. Starting or returning to school was described as a

significant event by the young people. School was highlighted as being important for re-establishing contact with friends and instilling a sense of normality into their lives.

Starting or returning to school also involved new challenges for the young people, their parents, carers, teachers and schools. Even if the young people, their parents and the education authority agreed on a suitable school, the head teacher could either be supportive or obstructive. Mary's mother found her 15 year old daughter's mainstream secondary school very good. Mary was the first pupil with a tracheostomy. She said:

'The school have done very well considering it was a first for them.'

In contrast, Jason said that he found his old mainstream secondary school obstructive:

'You can't go to the first floor ... so you can't go to school. Instead of saying... let's alter the classes and re-timetable.'

Jason was very unhappy with the suggestion that he should drop back a year or be transferred to a residential school for disabled children out of the borough. He did not consider himself disabled and opted to complete his GCEs with a personal tutor, then go straight onto 6th form. This worked very well and he felt that the 6th form college was far more progressive and flexible. For example, alterations were being made so that Jason could access the library. This positive attitude was very encouraging and he is now settled and studying for A levels. Jason then wants to study law at university.

Some parents said that their children had a lot of catching up to do. When Jessica started school she needed help with holding things such as a pencil and writing. Her parents and teachers did a lot of extra work with her and within a year she was making good progress.

Parents faced many dilemmas as to what was best for their children. One family had recently moved their 8 year old son from mainstream primary to a special school because they felt *'he was struggling'*. This had been a very difficult decision but seemed to be the right one because their son said he loved his new school. His mother explained:

'He used to get headaches and tummy aches and we haven't had anything at all (since he moved).'

Mohammed (aged 15) was studying for GCEs at a special school linked with his residential home. He felt education was extremely important and was looking forward to studying computing at college. He was absolutely adamant that any college he attended in the future must be for non-disabled and disabled people. Mohammed explained:

' Hopefully I can get a good job and earn plenty of money.'

He was asked, if given the choice, would he have chosen to go to a mainstream school as well? He paused and thought for some time. He then said:

' Yes, I suppose so.'

Some young people and their parents worked very hard to secure a place in a mainstream school. Parents said that they frequently rang the local education authority and asked to speak with people in authority. Success primarily depended on the flexibility and accessibility of schools in the area and the ability of parents to fight on their child's behalf. Ajaz (aged 6 at the time) initially went on a part-time basis to the local hospital school after discharge home. This gave his parents an opportunity to find an appropriate mainstream school. The one they selected was 15 miles across a major town. His father said:

'There was not much choice. The only reason is that it is all on one level, they have no steps.'

Ajaz did not experience any problems once he settled down. His parents drive over 300 miles a week taking Ajaz to and from school.

Support in the classroom

A number of things needed to come together to enable the young person to gain maximum benefit from starting or returning to school. Most, but not all the young people had a designated adult to look after them in the classroom. This was usually a member of their own care team and/or a classroom assistant provided by the local education authority. Jessica (aged 6) attended mainstream primary school and did not have any additional support. She spent all day with the same teacher and class. Her parents were not sure what would happen when she moved onto a larger secondary school and felt this situation would require reviewing.

Almost all young people with complex needs encountered much inflexibility with some of their carers in the classroom. The young people who were not able to take notes found their carers unwilling to do this for them. ***'It's not in the job description'*** was said to be a typical response by carers. This was particularly frustrating when taking notes was a necessary part of the classroom exercise. The young people said that was absolutely essential that they received appropriate educational support in the classroom that met their needs.

It was very important that young people got on and felt comfortable with their classroom assistants. The young people and their parents on the whole were not involved in their recruitment or selection and they wanted this to change.

Parents had encountered major problems when carers and classroom assistants were off sick. This usually meant that their children were not able to attend school. The young people were very unhappy if they had to stay at home unnecessarily.

Schools were generally said to have positive and supportive attitudes towards examinations. These could be taken orally, with the aid of a computer or by dictation. Some young people were gearing themselves up to prepare for sitting

examinations in this way. Jason had already achieved great success in his GCEs and was used to dictating for 3 hours at a time.

Break time and after school activities

Break time for some young people could be daunting. Parents said that a number of excuses would be used by their children to avoid going out into the play ground. The young people themselves talked about a number of reasons as to why things could be difficult. Some said they lacked confidence, whilst others found that specific things really bugged them. Mary (aged 15) found that new pupils stared at her tracheostomy. She said:

***'With the new intakes you get people staring at you...
sometimes when they ask me what it is.'***

Things improved as new pupils got to know her, but she experienced the same problem every new intake. Mary did not believe she was respected and sometimes had to cope with feeling different to everyone else. She was also very conscious of how other pupils viewed her with a minder in tow. She explained:

***'I would rather walk with her than have her follow me,
otherwise it's like being a dog on a lead.'***

Mary and her mother had been working hard with the school to allow her more independence. They had come up with the idea of issuing Mary with a walkie-talkie during breaks. This working well and she felt far more confident. Other children were also including her in activities as she no longer had an adult with her.

Not all schools provided a suitable range of clubs and activities. Ajaz said he would prefer more clubs for children like him as he cannot play sport. Mainstream schools were said to have clubs and teams events that concentrated on physical rather than intellectual or fun pursuits. The young people said they wanted and needed to feel that they are at the centre of the school community and not on the periphery.

Quality of lived experience, sub-theme 11: Aspirations for the future

The children and young people had a lot to say about growing up. They talked about their hopes and aspirations for the future. This included wanting to make more decisions for themselves and to be independent. Having friendships and relationships, especially sexual relationships were very important. The young people identified many barriers that prevented them from achieving these goals. There were several issues that these young people had to deal with in addition to the usual pressures of growing up. Many of the young people did not consider themselves to be disabled. They disliked the way society labelled and boxed them. Several young people wanted to make more non-disabled friends and were unhappy that their social lives revolved around groups for disabled people. Being independent was very important and trying to achieve independence presented many challenges. Very few said they had a role model or a person to aspire to. The young people looked to the future whereas their parents felt that it was difficult planning more than a few weeks ahead. Parents said they found letting go very difficult, especially as they had been used to fighting on their child's behalf. Some young people would still need support and protection as they grew up. Parents felt that getting the right balance was not easy.

Some young people had thought carefully about the future and had specific aspirations or careers in mind. Some teenagers had bank and savings accounts and were beginning to take responsibility for buying clothes and saving for more expensive items. Individuals had high expectations of what they wanted out of life. Jason explained:

'This is the life I want.'

He went on to suggest that agencies (such as health, social services and employment) would have to work around what he wanted rather than him work around them. This would require a change of mindset for many bureaucratic organisations.

Making decisions

The young people said it was very important to have freedom of speech and be listened to. Jason made a typical point:

'It is nice if I can say what I want rather than have people say this is what you want. It is just depriving people of free speech.'

For some young people independence and making their own decisions could not come quickly enough. They wanted to be more involved in making decisions and to be taken seriously. Not all doctors were equipped with the appropriate interpersonal skills to speak with and treat young people courteously. Many funny impersonations of well meaning doctors getting it completely wrong were recorded onto tape during interviews. Although hilarious, a very serious message underpinned the humour. The young people did not like being mistreated in this way. They wanted to be listened to and involved in making decisions.

Not involving the young person could have very serious repercussions. One teenager was very angry that her hospital consultant had written to her school recommending that she saw a psychologist to help her through her parents' divorce. She felt this had been done without her permission and behind her back and did not want the school being involved in what she considered private business. This episode was said to have serious consequences in relation to her confidence and self-esteem.

Being informed and having appropriate information was considered by the young people to be essential. Amber (aged 11) outlined typical comments:

'I need to know everything that is happening and what is going on.'

Lisa (aged 12) was also adamant she wanted more involvement. She explained:

'I just want to get on with my life and do my own thing, that's what I want to do, I don't need anybody telling me what to do, I just don't want that.'

Social inclusion

The young people encountered considerable problems in trying to get out on their own. Many felt that they were excluded from activities because they required supervision. For some young people with complex needs this resulted in having few friends and life could be lonely at times. Mary's mother had asked social services if she could have somebody to take her daughter shopping. After several months social services had not managed to come up with anyone. Like many young people Mary had her own mobile phone and was very mature for her age, but because she had a tracheostomy her social life was limited. She explained:

'I've got to take my suction with me. There is a night club for under 16s but I wouldn't be able to go.'

Some young people had learning difficulties that made them vulnerable. Parents said there were not many groups or organisations that provided interesting and stimulating activities for their children. Other families found that supervised activities for the over 16s were scarce. Louisa enjoyed attending a women's group, however, the group would not accommodate her when she reached 16. This was frustrating for parents whose teenagers needed support to develop independence and confidence.

Sexuality

The teenagers talked about relationships as being very important. Some young people were concentrating on school work and not as yet ready for a relationship. Others had boyfriends and girlfriends in the past or expressed they would like to meet someone in the future. The language used was on occasions frank and sexually explicit. Not everyone was happy about being '***young, ventilator-dependent and single***'. Some young people were adamant they wanted non-disabled partners. They said that their partners should not be expected to care for them as well. One teenager complained bitterly about his chances of meeting a girl. He said:

'Well you can't really have a girlfriend if you have got nurses pulling you along... you know what I mean! You know that chance is about one in a million!'

Wanting to give your carers the slip and have opportunities to meet people and spend time alone was a big issue for some. A number of young people had an intercom system wired up to their bedrooms. This had been used as a safety measure but was increasingly viewed as an invasion of privacy. Carers could be inhibiting unless they were of a similar age and part of the crowd. This was the case for one 17 year old who explained that when a member of his care team was getting married a surprise stripper-gram was organised in the pub. This was very much a group venture and a great time was had by all.

Two girls said they had plans to have a baby. One 11 year old wanted a baby when she was 16. Whereas Louisa (aged 13), was thinking a bit further ahead:

' I will get a husband and get married when I am about 27 and have a baby.'

Typical comments expressed by the young people around relationships were drawn as a cartoon (Illustration 4.3).

Illustration 4.3. Cartoon depicting that young people said that it was difficult to meet and spend time with boyfriends or girlfriends when they were supervised constantly by a parent or carer.



Quality of lived experience, sub-theme 12: The consequences of being a 'looked after' child

The 3 young people who were 'looked after' by social services experienced social exclusion in many aspects of their lives. Awareness of this exclusion ranged from being oblivious and relatively passive to feeling considerable anger and frustration. The young people highlighted the need to live with or have more contact with families and the need to be more involved in making important decisions about their lives. The prospect of young people returning to live with their families appeared to have reduced over time. Of this group:

- 2 lived in residential communities for young disabled people;
- 1 young person had occupied a hospital bed for 2.5 years and was currently waiting for a foster family to be arranged, and
- 1 young person had occupied a hospital bed for her first 6 years of life and although not formally 'looked after' by social services, her welfare

was monitored under the Children Act. She was transferred home during the study.

The experiences of 'looked after' children

Mohammed and John (both teenagers) lived in residential communities for young disabled people. These communities provided high quality accommodation and a secure caring environment. These factors did not however compensate for having limited contact with their families.

John explained he had lived away from his family for as long as he could remember. His mum, step dad, and siblings lived 2 hours drive away and John liked to visit them every month at home. The frequency of visits depended on the availability of staff. He was accepting of his current situation and enjoyed the contact he had with his siblings. Ideally, he would like to live alone with a team of carers and be nearer to his family.

Mohammed had grown up abroad in an extended family and subsequently settled in England. He moved to his current community following an accident when he was 8 years old. His parents and siblings lived about 2 hours away, but Mohammed was unable to get into their flat as the lift was too small. When he visited his family they had to meet in a local park or café. As a result he had been home only once in the past 4 months.

Distance from his family and culture had a considerable impact, especially in relation to his first language. He said:

'I used to speak it fluent (1st language) but now I can't speak it, I can say the odd words but I can't make a conversation'.

When asked how this affected his relationship with his family. He replied:

'I can understand what they are saying sometimes, but not all the time... so I just shout to them to shut up...I tell my

***brother to say it in English, so they speak in English...
sometimes.'***

Mohammed clearly would have preferred to maintain fluency in his first language and be able to get inside his parent's flat. In the past he had expressed a wish to live with his family but this had not been possible. He felt he was excluded from participating in even basic things with his family and this made him feel very angry. When he was older he wanted to live with his brother in rented accommodation and be nearer his family home.

Andrew and Sarah had both experienced a number of hospital placements. These included intensive care, children's wards and a specific unit for young ventilator-dependent people who were waiting to go home. These placements varied in their suitability and none was considered ideal. Although much effort had been spent adapting hospital facilities to give a homely feel, they cannot provide a secure, stable home. Andrew and Sarah did not have a separate bedroom, living area, or en-suite accommodation for their families to stay. Lack of facilities also restricted the preparation and eating of family meals around a table. Lack of transport to get out and about was also noted to be a problem.

Hospital wards are not counted as residential placements and social services departments are not required to conduct inspections. Social services reviews of the young people were also said to not always occur as regularly as they should do. These meetings were frequently cancelled or rearranged and the reasons for this were not clear.

Andrew who was primary school age, had been in hospital since his accident. He had a large extended family that faced many challenges. Social services were currently arranging for him to be cared for by foster parents. Andrew was very excited about his new family but talked almost exclusively about his birth family and in particular his siblings. He related their ages and what presents he wanted to buy them. His carer pointed out afterwards that his siblings were older than Andrew remembered. He had recalled their ages when he last saw them. It was not known what arrangements had been put into place for him to

maintain contact with his birth family. He was clearly keen to keep in contact, but was not aware of where they all lived or with whom. Memories and vital demographic data of his birth family had not been collated into an official record for him; young people need something tangible to keep memories of family and childhood alive.

Sarah had grown up in hospital and now loved being at home with her family. When asked if she had 3 special wishes? She replied:

'To come home...To go out to the shops...To go out to the park...'

These are experiences that most other primary school children take for granted and would probably not be considered worthy of a 'special wish'. Growing up in hospital is not normal and can have an impact on their quality of life.

Social lives of 'looked after' children

This group of young people generally described being deprived of some things and overloaded with others. Small communities can be very isolating and the children said that they had few friends outside of school or their residential complex. One person did not consider the residents he was living with as friends. Getting out and about was difficult if transport and staff were not available. Mohammed and John expressed they would like to meet more people from outside their own communities.

Being able to talk and hold a rational conversation with others was said to be very important. This was not always easy for a number of reasons. One young person said of his community:

'There are many (people) who are very mixed up.'

Some of these young people experienced an outpouring of well meaning generosity. This was especially so at Christmas, when traditionally children, particularly in hospital, are delivered gifts. This was described in the following way:

'Christmas last year was obscene, there must have been 5 sack loads of presents (for each person) and that was nothing compared to what came round on Christmas Day.'

The symbolic meaning of presents from siblings and parents could be obscured as young people did not know who had given what. Parents were not sure if they could compete against this level of generosity especially as most were coping with very tight budgets. Brothers and sisters could also be very jealous if they only received modest family presents. Going home to celebrate religious festivals with families was said to be rarely possible as staffing levels would not allow. Hospitals were also said to be a magnet for celebrities who would regularly visit and sometimes bring photographers with them. Some young people said that they had appeared in newspaper and magazine articles with celebrities, which they seemed to enjoy, but did not appreciate that there may have been an element of exploitation involved.

Carers who are responsible for young people living apart from their families are in a very difficult position. They are not able to take the risks that parents are prepared to with their own children. Inevitably, tensions can develop. One young person bitterly complained that his carers insisted on being with him ***'even when I go to the bog'***. He felt this was an invasion of his right to privacy and did not care if it was to ensure his safety. There were rules about watching videos of an appropriate classification and one young person found himself having to make an appointment to see his doctor to get permission to drink alcohol on his 18th birthday. He said:

'We also have to get permission from the managers to say whether we can have it (alcohol) or not...that is silly isn't it!'

These experiences contrasted sharply with those of Jason aged 17, who lived at home with his family. He enjoyed going to the pub, having a drink, choosing his own entertainment and generally being 'one of the lads'.

Involvement in decision-making

Involvement in decision-making varied greatly. When one young person was asked how much he was involved in making decisions about his life he said:

'I'm not really'. He explained that in hospital doctors and nurses 'tell you a lot more when it is quiet...(and) sometimes they don't ask me.'

Young people in residential communities and hospitals said they did not have contact with a General Practitioner. All their routine medical care was provided ***'in house'***. This has currently not caused any problems, but individuals need to know that they are entitled to consult with a General Practitioner and discuss any aspect of their health in confidence.

Some young people mentioned meetings that were convened to talk about their futures. These were said to be a very good idea, but did not always meet their needs in practice. One young person said:

'They don't always listen to me...they just turn round and say I think (name) says this and (name) says that... err I'm like errr sorry... I can speak for myself thank you!'

One young person was not happy with the way fellow residents were treated by staff. He had highlighted some of his concerns at a resident's meeting. He felt strongly that they did not have sufficient freedom or choices regarding their care and routines. He said:

' When I first came here ... if the staff wanted them (fellow residents) to go to bed, then they would have to go. It was just not giving them freedom...or choices... so I tried to change them (staff) ... so I asked them to change staff to give me some freedom.'

When asked if anything had changed? He replied:

'No not really... they are getting better...but because I was only 14 and coming in here making a big argument about giving them choices and all that ... some staff were like ... he's only a 14 year old... don't listen to him... he doesn't know.'

Being able to communicate articulately on behalf of those that are not so able, can be an enormous burden for a young person to cope with.

Both Mohammed and John said they would be sad when the time came for them to leave their residential communities. This would indicate that living in this type of accommodation met at least some of their needs. Mohammed and John said they were not able to go home and stay during school breaks or join their families for holidays. Both expressed a wish to have more contact with their families and especially their siblings.

Overall, these young people have had very little control over wider issues such as where they lived and with whom. The 1989 Children Act outlines that local authorities should provide Independent Visitors for children who do not live with their families. None of these young people said they had an Independent Visitor or an advocate independent of the system to act on their behalf. The young people who lived apart from their families generally appeared less likely to be successfully reintegrated as time passed.

Good Practice

Young people talked very highly of some key workers or nurses allocated to look after them. Having someone you trusted and to talk with was considered important.

Two young people mentioned they had a child protection charter, residents meetings and regular 'future meetings'. Although one person did not think they were always listened to, being able to voice concerns was considered vital. Being enabled to have some control over daily events in their lives was much appreciated and was considered a right.

Andrew was being transferred to the care of foster parents. This would enable him to live in a home surrounded by a foster family. He was very excited about this prospect and had enjoyed choosing things for his new bedroom. When young people are unable to live with their families, fostering is clearly a more appropriate option to living in hospital.

4.4 Discussion

The aim of this aspect of the qualitative study was to describe the health and quality of life of children and young people dependent on ventilators. This is the first major study undertaken with children and young people dependent on ventilators and findings described here provide a very rich description of how the children viewed their health and quality of life. These findings make an original contribution to the evidence base concerning the health-related quality of life of children and young people who are dependent on ventilators and their families.

Like the young adults in Brookes *et al.*'s study (2002) all of the children and young people said that the ventilator made them feel better as they expended less energy in trying to breathe adequately. The effect of the ventilator was to increase energy and their feeling of wellness, which enabled them to do things that they enjoyed. Health was a relative thing to each child and unless the child was older and had recently started long-term ventilation due to an accident or illness the children did not appear to assess their health in relation to others. This finding was also consistent with the parallel study conducted by the author with Heaton *et al.* (2003) into new uses of old technologies.

Constructs around health were not necessarily linked with the child's level of impairment. Indeed, for children and young people who had been dependent on ventilation since infancy, physical functioning did not feature in their conversations. Other issues such as dependency on the ventilator, length of time on the ventilator and the reason for ventilation appeared to impact more on the child's ability to engage in activities and quality of life.

Unlike the young adults in Brookes' study, the children and young people did not think about the technology and how it interacted with their body. This indicates that the thought processes of children and how they relate to the ventilator may be different to adults.

The children and young people described not being treated with respect and the discrimination they faced in trying to access appropriate services. The young adults in Brookes *et al.*'s study called this stigmatisation, which is a more sophisticated way of describing the same thing.

For the children and young people in this study, quality of life was clearly associated with quality of life experience, including: being treated with respect; being able to communicate effectively; being able to live at home in quality housing; receiving quality services; being able to get out and about and take holidays; having a good social life; receiving a good education; and being able to make decisions and gain independence. Issues around the availability of services and integration of the child into the family are similar to those described in Baumgardner and Burta's small study with parents (1998).

Ventilator-dependent children described clearly that their health and quality of life was better if they could live at home with their families and findings add weight to earlier studies on the potential damage that prolonged hospitalisation can have on the relationship between mother and child (Gross, 1984). The need to be able to communicate effectively emerged as an issue that had a profound negative impact on quality of life if children could not converse or engage socially with others; inability to communicate was found to be one of the most disabling barriers to social inclusion.

Findings illuminated the negative impact that professionals and poor quality services could have on the entire family, especially concerning lack of space in domestic homes, not being able to negotiate appropriate boundaries and losing control of decision-making concerning the family. Findings add weight to a small contemporary literature on the home as a place of employment and family

experiences of receiving care in their home (Coffman, 1995; O'Brien, 2001; O'Brien and Wegner 2002).

Some parents and children found the paternalistic attitudes of professionals to risk management very stressful to deal with and such issues are not discussed in the wider literature. To some extent, it is understandable that the natural traditions of doctors and nurses may be strongly 'protectionist'. Longitudinal studies of medical outcomes in the literature confirm that children on ventilators have relatively low rates of morbidity and mortality (for example, Frates *et al.* 1985; Canlas-Yamsuan *et al.* 1993), but nonetheless avoidable deaths did occur in far higher rates than in the population as a whole. For example, in their 10 year follow-up study of 39 children with spinal cord and neuromuscular conditions, Gilgoff and Gilgoff (2003) reported 8 deaths, of whom; 2 died following accidental ventilator disconnection; 1 died following a fall from a wheelchair; 2 were found dead in bed or their chair; and the remainder were due to illness or unknown.

Seven children in the current study said that they benefited from new and expensive technologies. These new technologies added significantly to the treatment and resource costs but were said to make children feel better, and had a positive impact on their quality of life. The cost implication of new technologies will be explored further in Chapters 5-7.

All of the things identified by the children and young people and their parents as contributing to their quality of life are arguably the same for all disabled and non-disabled children. The wider literature around disabled children generally has identified similar barriers that prevented disabled children from achieving quality life experiences (for example, Baldwin and Carlisle, 1994). The Audit Commission has undertaken contemporaneous evaluations of education and health services for disabled children generally and found similar inadequacies in health services, but problems were not reported on such a grand scale as the inadequacies experienced by some children in the current studies (Audit Commission, 2001, 2002a,b, 2003). This supports findings that ventilator-

dependent children have greater complexity of needs compared with the wider population of disabled children.

Findings suggest that the provision of a package of individually tailored high quality services and the removal of societal barriers to inclusion do make a positive impact to the quality of life of children dependent on ventilators. Unfortunately, not many of the children received all the services to meet their needs and/or services of a sufficient quality to enable them to enjoy life to their maximum potential. Providing the services to enable all children dependent on ventilators to live in optimum housing, enjoy being included socially and experience life to the full could have considerable cost implications; these costs will be explored further in Chapters 6 to 9. The inadequacies of the service provision for children dependent on ventilators has been highlighted previously in a public inquiry into the death of a child (Ashworth, 1996) and a subsequent report published by the Department of Health (1997a,b). The findings of this study confirm that inequalities and inefficiencies still exist in the health and social care provided for this group of children, which requires addressing.

The study described here was driven theoretically by an age appropriate health needs assessment framework (Pickin and St Leger, 1993) that was designed to identify if the needs of children were met or not. Whilst this is a technique commonly used in health services research to plan services and allocate resources, it caused a tension with some members of the advisory group who believed that provision of services should be allocated on a 'rights' based approach. It was thought that a needs-based approach afforded no guarantee in relation to allocation of resources, whereas as a rights-based approach could not be so easily contested once a legal precedence had been established. In the report of the initial exploratory study published by the Stationery Office (Noyes, 1999a), it was decided that the findings should be framed using Articles from the newly enacted Human Rights Act (1998). When framed in a legal rights-based context, it was demonstrated that a number of Articles had been breached. For example, a child has a right to live with and maintain contact with both his/her parents and lead a full and active life in society. This right was deemed to have been breached when the child spent prolonged periods of time

in hospital when they no longer had a medical need to be there (Articles 9, 23). A copy can be found in Appendix 2. This evidence has been used subsequently as evidence in a test case (*N v Department of Health in Northern Ireland*, 2001) brought by a parent of a child dependent on ventilator who believed that their right to a family life had been breached and in cases whereby children have sought compensation for their injuries and care following car accidents (Dyer, 2000). The implications of this will be discussed further in Chapter 10.

The views and experiences of professionals add an additional dimension to the perception and interpretation of health-related quality of life in this group of children. Doctors in particular experienced additional organisational and resource pressures as to whether to treat children or not and some viewed dependency on ventilation as a negative outcome. Findings concerning the pressures exerted by parents and advocacy groups to treat children have been discussed previously in Chapter 2. Professionals with more experience of working with ventilator-dependent children and their families in the long-term tended to be more positive about the benefits of long-term ventilation as an intervention.

Strengths and weaknesses

The major strength of this qualitative study was the fact that the research was primarily undertaken with children as opposed to on children (Ward, 1997). The design was child-centred, informed by a young ventilator-dependent person and methods of data collection were highly innovative and involved children who are not normally involved in research studies because of their perceived impairments. The research was also rigorously carried out, monitored by an advisory group and adhered to high ethical standards as outlined the National Children's Bureau (2003). Regular meetings were carried out during fieldwork and data analysis; throughout the study, the potential influence of the researcher was critically reflected upon and an audit trail was created outlining decision-making processes and outcomes. A number of transcripts as well as coding of the data were jointly reviewed as a means of enhancing theoretical

sensitivity, uncovering any biases and clarifying interpretation. Credibility was also enhanced by prolonged engagement in the field and by being advised by a young ventilator-dependent person. In addition, filed notes recorded the interview process, and together with interview transcripts and audio-tapes are available for scrutiny.

The Heideggerian phenomenological approach generated rich descriptions concerning how the children interpreted their health state, quality of life and quality of life experiences. The use of theoretical sampling maximised the range of information collected and the resultant sample which involved 67 parents and 53 children is substantial for a qualitative study of this nature. Saturation of themes was achieved and data were examined for rival explanations; negative cases were identified and examined to clarify the reasons for difference. The sample identified under-represented individuals such as minority ethnic families, 'looked after' children and fathers as being important to ensure the representativeness of the study. Recruitment was highly successful, mainly due to the excellent support of the hospital consultants who oversaw the process; over one third of the known population of children in the UK took part in this study. It is very unusual to be able to recruit so many minority ethnic families into a study such as this.

Following the initial exploratory qualitative study with 18 children and young people, a child-centred report was sent to children and their families for comment. Feedback strongly suggested that the interpretation of findings matched the views and experiences of interviewees thereby confirming the validity of the study. Findings from initial work reported here have been subject to peer review and subsequently published (Noyes, 1999a,b; 2000a,b,c; 2002).

Particular problems were encountered in trying to include 'looked after' children and young people. The study was successful in recruiting 3 'looked after' children and this is unusual as most studies exclude these children due to problems with access. Two young people were old enough to consent for themselves. In order to access a further child aged 9 years, it took 8 months of negotiation with the local social services department to get a signed consent

form before the child could be approached to see if he wanted to participate (which he did). Access to another child aged 6 years had to be abandoned after a long process which could not be concluded before the end of data collection. In this case the child's consultant had obtained permission for the foster mother to be contacted. The author obtained the consent of the local social services department, the child and foster mother, but as the parents had joint custody with social services, the local social worker insisted that parental consent was also obtained. The parents were in dispute with the local social services department and did not respond to any correspondence or telephone calls. Hence, it was not possible to interview this child. A phone call was received subsequently from the foster mother who said that the child who had not been in recent contact with his parents felt that his right to tell his story and be included in the study had been violated (which it had). This outcome was very unsatisfactory and guidelines need to be drawn up to guide researchers in such cases.

Methodologically, the main weakness of phase 2 of the data collection were that a number of elements (qualitative interview, quality of life questionnaire and collection of economic data) were packed into the time allocated to complete the case studies with the 35 children and their families. Unlike phase 1, it was not possible to visit the children on more than one occasion due to constraints on time and resources. More time would have allowed the children with communication impairments to explain their views and experiences in greater depth.

Strengths and weakness of the exploratory fieldwork undertaken with professionals has been discussed previously in Chapter 2. Synthesised findings derived from the 4 research themes will be reviewed in an evidence based and policy context in Chapter 9 and further research directions will be outlined in Chapter 10.

4.5 Summary

Chapter 4 continued the focus on the health-related quality of life of ventilator-dependent children (theme 2) by reviewing the few qualitative studies that have attempted to describe the health-related quality of life in ventilator-dependent children; these were found to be methodologically flawed and of poor quality. The chapter then reported a qualitative study underpinned by the social model of disability that aimed to explore how ventilator-dependent children viewed their health and quality of life and what impacts health, social and education services had on their quality of life. Proxy accounts of children's health and quality of life were also explored with parents. Findings provide new evidence around health and quality of life in ventilator-dependent children; children revealed that they defined their health in relative terms and perceived their health to be mainly good whilst parents' accounts tended to be more negative. Children and their parents reported significant organisational and societal barriers that had profound and negative impacts on the quality of life of their entire family. In contrast, many doctors had to deal with organisational and resource issues that may have had an influence on the way they perceived long-term ventilation as an intervention. Doctors with the most experience of caring for ventilator-dependent children and their families over a period of time tended to be more positive. The chapter concluded by comparing findings with published literature and discussing the strengths and weaknesses of the study.

The next chapter continues to focus on health-related quality of life in ventilator-dependent children (theme 2).

Chapter 5

Synthesis of quantitative and qualitative data around health-related quality of life

5.0 Introduction

Chapter 5 is the final chapter in the sequence that focuses on reporting empirical work around the health-related quality of life of children dependent on ventilators (theme 2). This chapter brings together quantitative findings from Chapter 3 and qualitative findings from Chapter 4. A narrative summary technique is used to bring together these data from different methodological paradigms to explore and explain similarities and differences around the measurement and experience of health-related quality of life of children dependent on ventilators.

5.1 Monitoring outcomes of ventilator-dependent children and young people

In Chapter 2, it was suggested that there are only 2 outcomes that really mattered to any paediatrician: the quantity of a child's life and the quality of a child's life (Eiser and Morse, 2001). The issue of life expectancy has not featured in the discussion thus far as it is not a focus of this thesis. However, in terms of completeness, it is important to mention that reports concerning mortality with this new and emerging group of children have concentrated on the monitoring of small numbers of cases managed in regional medical centres in the United States over relatively short periods of time (for example, Canlas-Yamsuan, 1992). The doctors who have published data concerning their own case loads acknowledge that this new population needs to be followed-up over a longer period of time in order to obtain more accurate estimates of morbidity and mortality.

Turning to health-related quality of life as an outcome, in Chapters 2 and 3, it was shown that searches of the qualitative and quantitative literature revealed a dearth of studies that explored and measured the health-related quality of life of children dependent on ventilators. The studies that have been published were located in different types of journals that were not readily accessible to, or accessed by, all professional groups involved in the care of ventilator-dependent children.

Having identified a significant gap in the evidence base around the health-related quality of life of this group of children, a programme of health-services research incorporating different methodological approaches was designed and carried out. The resultant qualitative and quantitative studies around health-related quality of life were reported in Chapters 2 and 3. Findings from these studies provided differing and sometimes opposing perspectives concerning the children's and their parent's perceptions of the child's health-related quality of life.

Exploratory fieldwork with doctors revealed that they associated more closely with quantitative outcomes and they were familiar with interpreting data derived from quantitative measures of health status and health-related quality of life. Most of the doctors interviewed had not read a qualitative study before and articulated their worries about the potential for bias in qualitative research. In contrast, parents, ventilator-dependent children and nurses said that they associated more with qualitative techniques and most had not read any studies incorporating quantitative outcomes. Nurses in particular said that they were not familiar with appraising the psychometric properties of a quantitative instrument.

Structured measures and ways of quantifying health status dominate outcomes research generally. The exploratory fieldwork identified that there was a potential danger that doctors would base their decision-making on the quantitative outcomes data alone which indicated that children dependent on ventilators had significantly lower health states than a population of school children. There was an obvious need to synthesise evidence from different methodological paradigms to ensure that qualitative and quantitative evidence around health-related quality of life was used to inform decision-making. It is the

aim of this chapter to try and synthesise findings in a meaningful way and methodological work had to be undertaken as part of the Fellowship to develop a way of achieving this end.

The need for a Cochrane Qualitative Research Methods Group

It has long been recognised that it is not possible for individual professionals and policy makers to search out and make sense of unmanageable amounts of information. The Cochrane Collaboration was founded to support health professionals by preparing, maintaining and disseminating systematic reviews of the effects of healthcare. The findings of high quality randomised controlled trials are central to the endeavours of the Cochrane Collaboration. However, it was recognised by like-minded researchers that findings from qualitative studies that assessed health interventions and outcomes could add value to quantitative evidence of effectiveness. It was also considered to be of benefit if evidence derived from different methodological paradigms and published in diverse journals that were not available or accessed by professionals could be brought together in a single review.

To this end, in 2003 the author co-founded the Cochrane Qualitative Research Methods Group to develop methods to include evidence from qualitative studies into systematic reviews. Evidence synthesis is one of the major areas of interest and methodological work is being undertaken on the processes by which findings from studies using different qualitative methods and generating a range of types of data can be synthesised and/or summarised and combined with quantitative findings. One of the methods that is currently being refined by researchers funded by the Economic and Social Research Council Research Methods Programme is the narrative summary technique.

5.2 Narrative summary as a method for synthesis

The narrative summary technique is primarily a qualitative and interpretative approach that has been developed as a way of juxtaposing qualitative and quantitative data (Denzin and Riessman, 1995; Dixon-Woods *et al.* 2004). The technique is flexible, allows the reviewer to be reflexive and critical, and can encourage insightful, original and vivid ways of seeing things. The major

advantage is that it is possible for qualitative and quantitative evidence to be discussed in parallel, if not fully integrated ways (Evans, 2002).

A narrative summary technique will be used initially to bring together data from different methodological paradigms to explore and explain similarities and differences in the quantitative and qualitative findings derived from selected individual case studies. Second, a narrative summary technique will be used to juxtapose qualitative and quantitative findings to explore and explain the outcomes for the entire group of children.

Case studies

The similarities and differences between qualitative and quantitative outcomes data concerning health-related quality of life in individual children are best illustrated through aligning, in parallel, qualitative and quantitative data derived from individual case studies. The qualitative data can then be used to explain and explore quantitative findings in an interpretive way. Data derived from 3 case studies are synthesised here.

Case Study 1: Jade

This study involves Jade a teenage girl who uses mask ventilation at night and lives at home with her family. Quantitative outcomes derived from the KINDL and EQ-5D are shown in Figure 5.1. On the KINDL, Jade's Total Quality of Life score and 5 of the 6 domains making up the total score are not significantly different to school children of a similar age, or her parents. KINDL scores correlated well with qualitative data whereby Jade described that she felt well, enjoyed school, liked where she was living, had good friends and engaged in a hectic social life. The qualitative data provided additional information around the fact that Jade did not require employed carers to look after her and she was looking forward to going to college, gaining a professional qualification and entering the world of work; these aspects were considered important by Jade but not measured on either of the quantitative instruments.

Jade had a medical condition that was associated with significant co-morbidity, but her score attributed to the Disease module was 100 (the maximum)

indicating that she was not unduly worried about her health; this was backed up in her interview.

Jade's score on the domain of Self-esteem was the lowest recorded of all the teenagers and significantly lower than school children and her parents. During the interview Jade said that she was fed up at the way she was treated and felt that as a person who was dependent on a ventilator she was not valued or respected by nurses, doctors and society generally (although she did not encounter problems with her close non-disabled friends). The KINDL picked up Jade's low self-esteem, but not the reasons that were arguably beyond Jade's control.

In relation to the EQ-5D, Jade recorded severe problems with mobility but no problems on the other domains. This resulted in her Health State score being significantly lower than school children, although during the interview she did not perceive her mobility to be a problem that stopped her getting out and about. The only issue that bothered her was the inadequate battery life on her wheelchair, which restricted the places that she could get to independently. As Jade was not likely to recover her ability to walk, she would not be able to increase her low Health State score on the EQ-5D. In contrast to her low Health State score, on the visual analogue scale Jade scored 100 for both 'health' and 'quality of life' (the highest score indicating she had the best possible health and quality of life). At interview, Jade said that at the present time she was very healthy and she clearly saw health as a relative state defined by her own experiences of feeling well and ill. If used as an isolated outcome measure, the Health State score could be interpreted by doctors and commissioners of services as an indicator that Jade experienced worse health and quality of life than school children, which was not borne out by the qualitative data.

Jade's parents had a different perspective on 3 out of the 5 EQ-5D domains and indicated that she had more problems than Jade herself perceived. This reinforces the importance of not relying on proxy data, which attributed a lower Health State score than Jade herself recorded.

Figure 5.1. Case 1 Jade: Quantitative outcomes.

Case 1: Teenage female living at home. Uses mask ventilation at night.			
KINDL	Child	Normative reference data: school girls	Parent
Total Quality of Life	68.75	70.78	80.21
Physical wellbeing	93.75	68.24	75.00
Emotional wellbeing	93.75	79.41	87.50
Self-Esteem	12.50	58.14	100.00
Family	68.75	75.51	56.25
Friends	81.25	78.06	81.25
School	62.50	65.19	81.25
Disease	100	78.67	87.50
EQ-5D			
Mobility	Severe problems	No girls age 15 had severe problems	Severe problems
Self care	No problems	99.9% of girls age 15 had no problems	No problems
Usual activities	No problems	95.8% of girls age 15 had no problems	Some problems
Pain/discomfort	No problems	73.3% of girls had no problems	Some problems
Anxiety/depression	No problems	58.9% of girls had no problems	Some problems
Health	100	79.55	100
Quality of life	100	NA	90
Health Index	0.34	0.87	0.30

Case Study 2: Krishna

This study involves Krishna, a teenage boy who was dependent on ventilation 24 hours a day and lived in hospital. Quantitative outcomes derived from the KINDL and EQ-5D are shown in Figure 5.2. Krishna's scores on both the KINDL and EQ-5D are significantly lower than school boys of the same age. In particular he failed to score any points on the domains of Friends and Self-esteem which may lead to the conclusion that this child did not value himself and had no friends. His parent's scores indicated some differences in perception, but mirrored a general trend concerning the low scores awarded by Krishna.

The qualitative data offered a detailed explanation as to why Krishna's scores were significantly lower than boys of a similar age and portrayed the child in a different light. Krishna said that in the recent past he had been involved in an accident, which resulted in him being paralysed from the neck down and dependent on a ventilator all the time. Krishna compared his health and quality of life at the time of data collection with his situation prior to the accident. Although he no longer needed to be in hospital, he was unable to go home as a package of care could not be organised and there were disputes about funding.

This meant that he regularly picked up chest infections in hospital, was not in contact with his friends, lived away from his family and was worried about his future health. Although Krishna did not indicate on the EQ-5D or KINDL, it was apparent during the interview that his mood was depressed and he was unhappy at his situation. The KINDL picked up his low self-esteem, but when probed he said that he did not feel that professionals valued him as a person and said that he desperately wanted to go home to get away from their negativity. Krishna felt that his health and quality of life would be much better at home and he was very upset that he was stuck in hospital and unable to take control of his life. He was keen to get back to his school and continue with his examination courses.

In this case it was essential to have both the qualitative and quantitative data in parallel in order to make a judgement about Krishna's health and quality of life. If the quantitative outcomes are taken in isolation, doctors and commissioners of services may come to their own conclusion that Krishna's life was not worth living and might not feel obliged to fund an expensive package of care to enable him to live at home, when in fact Krishna intimated that his life was worth living if only he could get out of hospital and get on with his life. The qualitative data highlighted the myriad of societal prejudices and barriers that were beyond Krishna's control and had a significant and negative impact of his health and quality of life.

Figure 5.2. Case 2 Krishna: Quantitative outcomes.

Case 2: Teenage boy living in hospital. Uses ventilation 24 hours per day.			
KINDL	Child	Normative reference data: Teenage boys	Parent
Total Quality of Life	43.75	73.53	44.79
Physical wellbeing	50.00	77.18	43.75
Emotional wellbeing	68.75	79.49	56.25
Self-Esteem	0.00	63.27	43.75
Family	68.75	79.56	6.25
Friends	0.00	78.43	0.00
School	75.00	63.58	68.75
Disease	37.50	78.67	25.00
EQ-5D			
Mobility	Severe problems	0.4% of schoolboys age 14yrs reported severe problems	Severe problems
Self care	Severe problems	No schoolboys age 14yrs reported severe problems	Severe problems
Usual activities	Some problems	7.4% of schoolboys age 14yrs reported some problems	Severe problems
Pain/discomfort	No problems	78.0% of schoolboys age 14yrs reported no problems	Some problems
Anxiety/depression	No problems	79.3% of schoolboys age 14yrs reported no problems	Some problems
Health	30	83.94	33
Quality of life	40	N/A	33
Health Index	0.09	0.90	-0.17

Case Study 3: Sam

This study involves Sam, a young schoolboy who was dependent on ventilation 24 hours a day and lived at home. Quantitative outcomes derived from the KINDL and EQ-5D are shown in Figure 5.3. Sam had profound intellectual and physical impairments so his mother provided proxy estimations of his health and quality of life. His mother's scores on the KINDL and EQ-5D revealed that Sam had lower Total Quality of Life and Health State scores than schoolboys of the same age. The qualitative interview revealed that at the time of data collection, Sam was not his usual self, had an infection, and was not able to go to school. These factors meant that his mother scored her child lower on the domains of Physical wellbeing and Disease. His mother said that she would have scored differently if he was well and this would have resulted in a KINDL Total Quality of Life score in the same range as schoolboys of a similar age.

The qualitative data revealed that Sam's mum described his health in relative terms and did not compare her child with other non-disabled children of the same age. When Sam was well, she said that she would have given him a higher visual analogue score as opposed to the 70 she awarded when he was

unwell and off school. At interview, Sam was observed to have impairments that were on the most profound end of the spectrum of all the children included in the study. His mother said that he was much loved and together with his full-time team of employed carers, he enjoyed a good quality of life. His parents and employed carers were adept at interpreting his individual ways of communicating and apart from being unwell, he seemed contented and settled. Much of the house had been converted to meet Sam's needs and his family focused their lives around making his life as enjoyable and comfortable as possible. Concerns around using generic tools with profoundly disabled children will be discussed later in the chapter.

Figure 5.3. Case 3 Sam: Quantitative outcomes.

Case 3: Young school boy living at home. Uses ventilation via a tracheostomy 24 hours a day.			
KINDL	Child N/A	Normative reference data: School boys	Parent
Total Quality of Life		76.67	60.71
Physical wellbeing		76.68	18.75
Emotional wellbeing		82.89	56.25
Self-Esteem		66.52	43.75
Family		83.58	87.5
Friends		78.21	75.00
School		72.35	Not in school at time of data collection
Disease		78.67	50.00
EQ-5D			
Mobility		98.1% of boys age 7 had no problems	Severe problems
Self care		90.2% of boys age 7 had no problems	Severe problems
Usual activities		98.1% of boys age 7 had no problems	Severe problems
Pain/discomfort		92.5% of boys age 7 had no problems	Some problems
Anxiety/depression		80.4% of boys age 7 had no problems	Some problems
Health		94.10	70
Quality of life		N/A	84
Health Index		0.91	-0.17

Synthesis of qualitative and quantitative outcomes for the entire group

A narrative synthesis of qualitative findings derived from interviews and observations with 50 mothers, 17 fathers and 53 children and young people (Chapter 4) will be juxtaposed against the quantitative data derived from the entire group of 17 children and 28 parents (Chapter 3).

Statistical difference and association versus rich description and explanation

From a statistical viewpoint, the analysis was interested in both group differences and the association between 2 variables such as children's and their parent's estimates of the child's quality of life on a visual analogue scale. Quantitative findings for all 17 children and young people derived from the KINDL and EQ-5D are summarised in Table 5.1.

The children and young people had significantly lower KINDL Total Quality of Life scores on 5 out of 7 domains (except Friends and Disease) than a population of school children.

Parallel qualitative findings revealed that despite feeling healthy generally, the children still got breathless and tired which meant that their Physical wellbeing scores were significantly lower than school children even if they did not feel ill or were not in pain. The ventilation made children feel better and gave them more energy, but these states were seen as relative and most children and parents did not compare themselves with other non-disabled children of a similar age. Children who had recently been injured and could remember their pre-injury state did make an assessment of their situation relative to their pre-disabled state and generally gave themselves lower scores.

Table 5.1. Summary of mean differences for all children dependent on ventilators compared with school children and parents.

	Mean differences: all 17 ventilator-dependent children compared with school children	Mean differences: all 17 ventilator-dependent children matched with their parents	Mean differences: 17 ventilator-dependent children compared with 28 parents
KINDL			
Total Quality of Life Score	Ventilator-dependent children had significantly lower scores P=0.0009 (-20.23 to -8.46)	Correlation r=0.70 P=0.001	Correlation r= 0.70 P=0.001
Physical wellbeing	Ventilator-dependent children had significantly lower scores P=0.004 (-19.86 to -4.86)	Correlation r=0.53 P=0.03	Correlation r=0.53 P=0.03
Emotional wellbeing	Ventilator-dependent children had significantly lower scores P=0.014 (-17.62 to -2.29)	Correlation r=0.52 P=0.03	Correlation r=0.52 P=0.003 Parents scored significantly lower than children P=0.03 (-14.13 to - 0.46)
Self-Esteem	Ventilator-dependent children had significantly lower scores P=0.005 (-35.12 to -7.44)	NS	Parents scored significantly higher than children P=0.002 (5.90 – 23.84)
Family	Ventilator-dependent children had significantly lower scores P=0.006 (-26.61 to -5.4)	Correlation r=0.78 P=0.001	Correlation r=0.78 P=0.001
Friends	NS	Correlation r=0.84 P=0.0004 Parents had significantly lower scores than children P=0.04 (-16.24 to -0.16)	Correlation r=0.84 P=0.00004
School	Ventilator-dependent children had significantly lower scores P=0.008 (-21.96 to -4.1)	NS	NS
Disease	NS	Correlation r=0.51 P=0.03 Parents had significantly lower scores than children P=0.007 (-27.48 to -5.16)	Correlation r=0.51 P=0.03 Parents scored significantly lower than children P=0.007 (-24.40 to - 8.10)
EQ-5D			
VAS health	There was no significant difference between ventilator-dependent children (boys or girls, or between age groups) and school children	NS	NS
VAS Quality of Life	N/A	Correlation r=0.67 P=0.003 Parents had significantly lower scores than children P=0.02 (-17.79 to -1.64)	Correlation r=0.67 P=0.003 Parents had significantly lower scores than children P=0.02 (-17.71 to -1.64)
Health state	Ventilator-dependent children (boys and girls and both age groups) had significantly lower scores than school children P=0.000001 (-0.74 to -0.36)	Correlation r=0.58 P=0.01 Parents had significantly lower scores than children P=0.02 (-0.32 to -0.02)	Correlation r= 0.58 P=0.01 Parents had significantly lower scores than children P=0.02 (-0.32 to -0.02)

Emotional wellbeing scores for the group as a whole were significantly lower than for school children and parents' scores were significantly lower than childrens' scores. The qualitative data revealed that this was a heterogeneous group and the only common factor was that everyone used long-term ventilation (albeit different types and with different levels of dependency). Children were at various stages of rehabilitation and some of the older children were living with life-limiting diseases. Individual circumstances shaped the children's perceptions regarding their emotional wellbeing, which varied on a continuum. For example, the children who lived in hospitals said that they were bored, and felt alone and were insecure because hospitals were totally unsuitable places to

live, whereas other children and young people who lived at home were socially included and said they enjoyed varied life experiences. The aggregated KINDL Emotional wellbeing score does not reflect these individual differences. The qualitative data revealed that many of the issues around emotional wellbeing depended on such things as quality housing, transport and access to good quality and appropriate services. Most of these issues were beyond the control of the child and yet impacted significantly on their wellbeing and quality of life generally. Parents appeared more aware of the global effects of these issues on the child and their lower scores reflected these concerns.

Qualitative and quantitative findings around self-esteem confirm that children and young people have significant problems. Children reported on the KINDL that they did not always feel proud, pleased with themselves or on top of the world. Whereas in the qualitative interviews, children said very clearly that they did not like the way that they were treated by professionals and society in general and felt that professionals did not always value them as people. Parents' scores were significantly higher than their children and interviews with parents did not reveal concerns about their children's self-esteem.

The domain concerning the Family was significantly lower for children dependent on ventilators compared with school children. This domain looked at various aspects concerning relationships with parents. The qualitative data revealed that a high proportion of the children had spent significant periods of time in hospitals and this could have a negative impact on relationships. Many of the children and young people said that they got on well with their parents but it was their employed carers and professionals that disrupted and restricted their lives. This aspect is not covered by the KINDL.

Children and young people dependent on ventilators did not have significantly different scores on the domain of Friends than school children. This aggregated score does not reflect the varied experiences of children within this group; some children had good social lives whilst others did not. Qualitative interviews revealed that some children were desperately lonely and isolated from their friends (especially those stuck in hospital). In contrast, children who had access to good transport and the ability to get out had lots of friends and a

varied social life. The children with good friendship networks tended to be those who were not dependent on ventilation 24 hours per day. One of the biggest issues for the young people was not just the ability to have 'friends', but the desire to have close relationships, and the teenagers talked about sexual relationships. This issue is not covered in the KINDL and yet was very important to teenagers.

On the domain of School, children dependent on ventilators scored significantly lower than school children. Children identified problems around completing schoolwork and getting bad marks or grades. The qualitative data helped explain why this should be so. Children who lived in hospital said they missed out on their formal education as hospital teachers were not equipped to deliver the national curriculum. Not all children had been enabled to communicate and they could not interact with the teacher or class. Choices regarding schools were said to be limited and not all children had the support they required in the classroom. Many children missed significant periods of school because their carers were off sick and the school would not accept them without support. Children also missed out on school because they were at times unwell. Many of these issues were beyond the control of the child and were more to do with barriers that prevented them from receiving quality education.

On the domain of Disease children did not have significantly different scores than a population of children with chronic illnesses, but parents scored significantly lower than their children. The parallel qualitative data revealed a mixed picture. Some children were afraid that their health might get worse but others were not. Being afraid tended to correspond with those children who had life-limiting diseases. Almost all of the children missed something at school or college because of their health (KINDL), but this was just as likely to be caused by lack of employed carers as ill health in the child.

A confusing picture emerged with the EQ-5D whereby the children and young people had significantly lower health states than a population of school children but there was no significant difference concerning self-rated health on the visual analogue scale. Parallel findings from the qualitative study revealed that children considered their functional status to be less important than their ability

to enjoy quality life experiences. Children and young people who had adjusted to their long-term ventilation clearly judged their health in relative terms as to how they were feeling and did not tend to compare their health with other non-disabled children. Only the group of children who had recently been injured measured their health against their pre-injury state. Synthesis of findings across studies confirm that in relation to ventilator-dependent children the self-rated measurement of health on the visual analogue scale was a more appropriate approximation of their overall self-rated health-related quality of life than the aggregated Health State score which incorporated levels of physical functioning. Parallel qualitative data was also able to provide a rich description that helped to explain why there was a strong and positive relationship, and significant difference between children's and parent's self-rated quality of life scores recorded on the visual analogue scale. Qualitative findings indicated that the judgements children and their parents made about quality of life were different and dependent on individual constructs as to what constituted a good quality of life. Children viewed their quality of life relative to their own life experiences, whereas parents tended to compare children's lives with their own non-disabled life and as such ascribed lower scores than children.

The health-related quality of life of children in hospital versus children at home

The synthesis of qualitative and quantitative findings across studies provide consistent evidence that children living in hospital experience fewer positive life experiences and report their health-related quality as being lower than children living at home. Although only 2 children living in hospital were able to complete questionnaires to measure their health-related quality of life, the accompanying parallel qualitative interviews revealed rich descriptions concerning the extremely limited life experiences of children living in hospital and their strong desire to live at home with their families.

5.3 Discussion

Philosophical and theoretical considerations

The qualitative and quantitative data synthesised in this chapter were derived from studies embedded in different philosophical paradigms and underpinned by different theoretical perspectives. The quantitative measurement of health-related quality of life was underpinned by the medical model of disability, which defined a 'normal' healthy child and measured negative deviance from the norm as an indication of decreasing health, quality of life and unmet 'need'. The significantly lower scores of children dependent on ventilators could be interpreted as a disbenefit of long-term ventilation as an intervention and may be used in the allocation (or restriction) of funding in a resource limited health service.

In contrast, qualitative interviews sought to define the children's own perceptions around health and quality of life, and was underpinned by the social model of disability, which conceptualised disability as a social and political issue rather than a medical one. The findings were then framed around the newly enacted Human Rights legislation (Appendix 2). Findings indicate that children conceptualised health and quality of life in their own ways, encountered many societal barriers and their rights in law were not always met. Across studies there was consensus that the self-esteem of ventilator-dependent children could be affected by the negative and disabling experiences children encountered. Findings add weight to the small literature concerning the evolution of emotional problems experienced by children and young people dependent on ventilators (Dalton and Kirkhart, 1985; Gipson *et al.* 1987; Fischer, 1989; Gorman *et al.* 1998).

Qualitative studies also revealed that some children demonstrated a high degree of resilience in relation to their circumstances. Lawford and Eiser (2001) have begun to explore the links between the concepts of quality of life and resilience, and believe there may be a relationship between individual variation in quality of life reports and the resilience of children; although they stress that further empirical work needs to be undertaken to develop a theoretic model.

Legal test cases outlined in Chapter 3 illustrated that parents were beginning to use the Human Rights Act (1998) to force the NHS to provide their children with long-term ventilation and expensive packages of care, irrespective of their child's health, quality of life or whether doctors thought they were likely to benefit from long-term ventilation. Using a legal route to secure treatment and funding appears to be in tension with principles of evidence base medicine whereby high quality evidence of effectiveness is used to underpin decision-making and the allocation of resources (Harris, 1987).

Synthesising the findings went some way to bringing these positions together by using the qualitative findings to explore and explain, in parallel, the quantitative findings. Quantitative measures were able to discriminate differences in the health and quality of life of ventilator-dependent children compared with school children and other groups of chronically ill children. Whereas qualitative findings offered some illumination as to why parents and ventilator-dependent children felt that their treatment was justified and highlighted the weaknesses in using traditional quantitative measures as a discrimination index to inform decision-making. When qualitative and quantitative findings were juxtaposed, it was clear from parents' and childrens' narratives that when health-related quality of life measures were used as a discriminative index, it was possible that lower scores could be confused with lower health and quality of life when this may not necessarily be the case.

Confusing functional disability with quality of life

The EQ-5D in particular, was found to be responsive in measuring the child's functional disabilities but the resultant Health State score was not necessarily a reflection of the child's health-related quality of life.

Given these findings, the negativity expressed by members of the advisory group towards the use of such measures appears understandable and explains why they wanted to see the qualitative findings framed around the Articles in the Human Rights Act (1998). A valuation of human beings according to their functional capacity could then be seen in sharp contrast to the humanistic values laid down in the Human Rights Act; hence parents have successfully

used the Act to secure resources for their children in situations where they had been previously denied resources from the NHS.

The disbenefits of hospitalisation on children's health-related quality of life

The studies reported here are important as they provide original evidence and add to the small evidence base around the contemporary experiences of children who live in hospital settings. Few studies have looked at the psychosocial situation or outcomes of children with long-term ventilation who live in intensive care units or hospital wards. One reason for this is that it is particularly difficult to obtain ethical approval to interview children whilst in hospital (Stalker *et al.* 2004).

The children and young people did not want to live in hospitals and recent test cases confirmed that such treatment constituted a violation of their right to a family life. Synthesis of findings across studies found that hospitalisation had a profound and negative impact on the health-related quality of life of children.

Apart from the current studies, there are a few notable exceptions whereby research has been carried out with ventilator-dependent children whilst in hospital. Effey (1999) conducted a quantitative time and motion study over a 2 day period into the duration and quality of social contacts of children living in an intensive care unit, and Schwerdt, (1996) presented experiences from his own intensive care unit. Findings confirm that children had little and poor quality contact with nurses and the significant people in their lives, such as their parents. Colville and Mok (2003) reported 2 cases of self-harm inflicted by children dependent on ventilators who lived in an intensive care unit. They concluded that boredom, lack of therapeutic care and total unsuitability of intensive care as a place to live contributed to the children's behaviour.

There is contemporary supporting evidence in the wider literature that children who live in hospitals are deprived of sleep (Corser, 1996), experience unnecessary painful procedures (Southall *et al.* 1993), experience disrupted and inadequate education (Bolton 1997), and suffer psychological

consequences (Tichy *et al.* 1988; Warner and Norwoord, 1991). All these reports add weight to the findings from the current studies that hospital, and in particular intensive care environments, are totally unsuitable places for children to grow up in and have a profound and negative impact on their quality of life.

Potential use of health-related quality of life measures as predictive or evaluative indices

In the current study health-related quality of life measures were used as a discriminatory index. However, both qualitative and quantitative findings confirmed that ventilator-dependent children suffered from low self-esteem in particular. Quantitative measures may therefore have potential use to predict or classify ventilator-dependent children with low self-esteem in order to provide appropriate therapeutic interventions aimed at raising their self-esteem. The KINDL may also be sensitive to measuring the magnitude of longitudinal change in response to such therapeutic interventions in an individual or group. Further work is needed to validate health-related quality of life measures as predictive or evaluative indices with this group of children.

Strengths and weaknesses

One of the weaknesses of the narrative summary technique as a method of synthesis is the potential lack of transparency and risk of bias. The technique is heavily dependent on the skills and prejudices of the researcher and one form of evidence or the other can end up as the poor relation. One of the perceived advantages of aligning qualitative and quantitative outcomes was that the qualitative data provided the context and rich background data concerning children and their circumstances. This qualitative data was very powerful and without careful consideration could be inadvertently used to play down the quantitative outcomes, which in some instances were statistically highly significant. One example of this could be seen with the children who had profound intellectual and physical impairments. Their parent's proxy quantitative estimates revealed significantly lower health states and quality of life, but the qualitative data indicated that parent's perceived their children to have a 'good' quality of life that was defined by them as opposed to society in general. Two

parents in the study had fought for their children to be offered long-term ventilation and it is difficult to determine whether this experience had affected their objectivity.

One of the strengths of this narrative synthesis was that it exposed a number of methodological concerns about the use of quantitative instruments with children with disabilities generally and in particular children with functional impairments. The qualitative methods found that the children defined their health and quality of life in different ways to the functional elements measured by the EQ-5D. For most children health and quality of life were seen as relative to them and meant different things to individual children and parents. The EQ-5D was originally designed for adults and was extrapolated for use with children. Although the instrument was validated with a population of school children, this was the first time it had been used with disabled children and findings question whether disabled children share the same views about the meaning or implications of functional disability.

Eiser and Morse (2001) did not consider disabled children specifically in their systematic review of quality of life measures in childhood and no instruments were identified that had been developed with children with existing physical and intellectual impairments. Some initial developmental work is currently being undertaken by a pan-European team to develop and test the first instrument for use with disabled children and adolescents (Bullinger *et al.* 2002). This instrument will be developed and tested with populations of disabled children (as opposed to non-disabled children) across Europe.

A group-specific health-related quality of life instrument has not been developed for children dependent on ventilators and is unlikely to be so as the population is too small to test psychometric properties. A more child-centred and disability-aware approach to the measurement and discrimination of health and quality of life of ventilator-dependent children (and disabled children generally), compared with 'normal' children, is needed to inform decision-making. Future studies may have greater validity if qualitative methods are incorporated and instruments designed specifically for disabled children are used instead of those that compare children with 'normal' populations of school children.

Synthesised findings derived from the 4 research themes will be reviewed in an evidence based and policy context in Chapter 9 and further research directions will be outlined in Chapter 10.

5.4 Summary

This chapter brought together quantitative and qualitative findings around the health-related quality of life of ventilator-dependent children (theme 2), derived from Chapters 3 and 4, by using a narrative summary technique.

Synthesis of findings across studies illuminated the differences between the measurement and experience of health-related quality of life as perceived by the children themselves, their parents and healthcare professionals. Qualitative data was used to explain the quantitative findings. Explicit linkages were made between how children viewed and experienced their health and quality of life, the inadequacies of generic measures to capture the children's actual experiences or the disabling barriers that impact on their quality of life, and the use of proxy estimates.

Qualitative and quantitative findings concurred that ventilator-dependent children experienced low self-esteem, and that generic measures may have some utility as an assessment tool and may be used to measure change over time.

The chapter concluded by discussing the strengths and weaknesses of the narrative summary technique as a method for synthesising quantitative and qualitative data.

The next chapter changes the focus of the thesis onto the costs of care (theme 3) and satisfaction with services (theme 4).

Chapter 6

Evaluating the cost of care and satisfaction with services using quantitative methods: How much does life on a ventilator cost?

6.0 Introduction

Chapters 6, 7, and 8 are linked sequentially and report empirical work around the costs of caring for children dependent on ventilators (theme 3), and satisfaction with services received (theme 4). In this chapter the costs of care and satisfaction with services are evaluated using quantitative methods that are underpinned by the medical model of disability. Chapter 7 explores and explains individual patient and family costs, and satisfaction with services using qualitative methods underpinned by the social model of disability. Then in Chapter 8 a narrative synthesis of quantitative and qualitative findings derived from Chapters 6 and 7 is presented.

Chapter 6 begins by exploring the distribution of resources through legal imperatives around the allocation of finite resources, and in particular via a test case whereby a mother of a child used the Human Rights Act to access resources to enable her child to live at home - a choice that had been previously denied by the NHS.

The chapter then goes on to discuss the economic evaluation of costs for this group of children and reviews the few quantitative studies that have been undertaken with children dependent on ventilators. A study evaluating the costs of care and services received in the previous 12 months by 35 children and young people dependent on ventilators is then reported.

6.1 Life as a ventilator-dependent child: A life worth paying for?

Two of the most frequently raised questions during exploratory fieldwork were *'is life on a ventilator worth paying for?'* and if it is worth paying for *'how much should the tax payer reasonably pay for it?'*

The debate around the costs of caring for children dependent on ventilators came to prominence in the UK following the publication of an article in the British Medical Journal entitled 'A Difficult Case: Home ventilation of a child with motor and sensory neuropathy' (Davies, 1996). The aim of this case study was to be controversial and provoke debate by highlighting the case of a young ventilator-dependent child whose 16 months of care at home was estimated to have cost the NHS £160,000 in 1994. The child had been managed in an adult intensive care unit and the only other alternative, apart from home care with a package of support services, was to equip and staff a room in the children's ward for her.

Davies (1996) argued that readers may consider that the money could have been better spent by relieving a greater quantity of misery, but applauded the decision of the health authority as *'a costly but beautiful deed not susceptible to financial analysis'*.

A debate ensued in the electronic response section of the British Medical Journal website. The main thread of this debate centred on the concept that *'life does have a price and the rest of us have to pay it'* (Hadden, 1996). The mother of the child who contributed to the article claimed that it was not possible to put a price on her child's life and yet Hadden (1996) and others who responded argued that a price was nevertheless paid by UK citizens through taxation. Hadden (1996) argued that there would become a point at which the morality of providing free care for ventilator-dependent children and their families would be exactly balanced by the immorality of taxing citizens so hard that they would have insufficient funds to meet their own needs.

The choice of long-term ventilation or a dignified death for patients is determined in part by the wealth of the patient and or the priorities and resources available within the healthcare system (Krishnan, 1993). In all developing and most middle-income countries long-term ventilation is not considered a viable economic option; patients are made comfortable with palliative care and the ventilator is withdrawn in a controlled way thereby allowing them to die (Gardner *et al.* 1985).

When decisions regarding the prioritisation of need and corresponding allocation of health expenditure have been put to the general public, different priorities have been reached than those taken by politicians and commissioners of services who traditionally allocate resources. One such example is the Oregon experiment in the United States (Rosenbaum, 1992). In 1989, faced with spiralling healthcare expenditure, the Oregon Health Services Commission was mandated to study prioritisation of healthcare resources as part of a package of bills. The resulting public consultation exercise afforded a low prioritisation to certain services for women of childbearing age and children; in particular neonatal intensive care for sick babies was given a low priority which would have led to considerable cuts in service. The very high cost and poor outcomes of these babies was reported as being the major influence on public opinion; in particular the high incidence of brain damage and need for long-term care in over a third of premature babies.

In the UK, the Courts have taken a particular view about the obligations of the state, for example, in the Diane Pretty test case (*Pretty v Department of Public Prosecutions*, 2001). The Court held that ***'the right to human dignity which is enshrined in Article 3 of the Human Rights Act is not the right to die with dignity but the right to live with as much dignity as can be afforded until life reaches its natural end'***. When this judgement was reached, it raised a lot of questions concerning whether this meant dignity at any financial cost and just how much dignity could be afforded when resources were finite?

Ventilator-dependent children: An automatic right to resources whatever the cost?

Over the last 3 decades, there has been considerable debate concerning the level of support available to disabled children and their families. Baldwin's groundbreaking work in the 1970s and 80s sought to explore the legitimacy of the need of families with disabled children for extra support from the state (Baldwin, 1985). At the time of Baldwin's study, screening techniques during pregnancy for foetal anomalies were less sophisticated and a high percentage of disabilities were not diagnosed until immediately or soon after birth. Many children with serious congenital anomalies did not survive the first year of life. Around the time of Baldwin's study there were a number of high profile cases whereby parents had either refused consent to have their disabled children treated which was subsequently over-ruled by the Courts (Williams, 1981), or whereby the paediatrician with the consent of the parents had sought to bring an end to the 'suffering' of the disabled child and had subsequently been charged with murder following intervention by a pro-life lobby group (Kennedy, 1982). Weighing up the evidence in the early 1980s, Baldwin concluded that most parents did not have a choice as to whether their disabled child was born or treated, as limits in the then screening technology and legal precedents determined treatment decisions. Thus, she concluded that as society had saddled parents with a considerable burden not always of their choosing, then society should provide extra welfare benefits and support in the form of targeted health and social services.

Since Baldwin's study in the late 1970s and early 1980s, there has inevitably been considerable evolution of both policy and practice. In the 1970s, it was still common practice for disabled children to live away from their families for all or part of the time in institutional care. Recent Government policy has closed almost all long-term institutions for disabled children under a strategy that aimed to improve the quality of life and outcomes of these children by transferring budgets from providing institutional care to enable disabled children to live with their families at home. Thus, as described in Chapters 1 and 2, at the same time that increasing numbers of severely disabled children were surviving with

greater complexity of need for health and social care, the options for their future placement have been limited (albeit for sound reasons) to an acute hospital bed, living with their family, or a substitute foster family.

Much health policy over the last twenty years has been about containing escalating costs and there has been an expectation that parents (especially mothers), should be responsible financially for their children, and shoulder the burden of caring for their disabled children at home with limited support from the state (Dobson and Middleton, 1998). As a result of these expectations, families of disabled children have been found to have reduced capacity to earn a living, increased costs of caring for their disabled children and are frequently socially isolated with a higher risk of family breakdown. The emergence of a new population of disabled children who are dependent on technology to survive, some of whom require 24 hour care, has increased the level of dependency of children to the point that it is not viable for most families to care for their children at home unless one or both parents give up work to become full-time carers or a package of state funded support is provided. Thus parents of children with the most complex needs are often left with no option other than to leave their child in hospital until sufficient money is available to purchase a package of care to enable the child to live at home.

The ethical positions around treatment of disabled children that were described previously in Chapters 2 and 3 have also evolved over the last 2 decades. Driven by the changing attitudes of society generally towards disability, there has been much work undertaken to determine the circumstances under which treatment may be withdrawn or withheld from children, culminating in published guidance for doctors (Royal College of Paediatric and Child Health, 1997). This guidance is currently being revised once again and reflects the impact of the Human Rights Act (1998), which incorporates the right to life and special rights of protection to disabled children.

Disputes concerning finite health resources

In cases of disputes about finite healthcare resources, the Court of Appeal issued guidance on the general principles to be applied in establishing priorities

in *R v North West Lancashire Health Authority* (2000). The appeal judges held that in establishing the respective needs of patients suffering from different illnesses and determining the respective strengths of their claims to treatment or services, it is vital for an Authority to:

- Accurately assess the nature and seriousness of each type of illness;
- Determine the effectiveness of various forms of treatment for it, and
- Give proper effect to that assessment and that determination in the formulation and application of its policy.

This judgement by the Court of Appeal reinforces the need for evidence based healthcare in determining the cost-effectiveness of healthcare interventions and the prioritisation of resources. As outlined previously in Chapter 1, there is a wealth of evidence derived from health technology assessments that the ventilator is an effective piece of equipment when used in the intensive care unit to maintain life in the short term; but there is little rigorous evidence about the efficacy of maintaining a child's life long-term on a ventilator in domestic settings or evaluation of their long-term outcomes. Nor is there much evidence on how health services for ventilator-dependent children can be managed, organised and delivered effectively in the community.

Whilst evidence on the costs and effectiveness of long-term ventilation in children have remained elusive, as explained in Chapters 2, 3 and 4, parents and campaigners in the UK have argued that access to long-term ventilation as an intervention should be the right of every child irrespective of their disability, postcode, arbitrary policies concerning eligibility criteria, and the ability of parents to fight for resources. Taking this argument to its logical conclusion, ventilator-dependent children should have a legitimate right to receive long-term ventilation and a package of support services regardless of finite resources or local priorities concerning the needs of the population as a whole. This was clearly the opinion of Davies (1996) who described the £160,000 cost of supporting a ventilator-dependent child at home for 16 months as '**a costly but beautiful deed not susceptible to financial analysis**'.

Test case

One parent has recently used the Human Rights Act (1998) to secure funding for her ventilator-dependent child to live at home. In 2001, the child's mother initiated a judicial review in Northern Ireland as her 15 month old child had been in hospital since birth because the Deputy Secretary of the Department of Health in Northern Ireland had refused to fund a package of home care on the grounds of cost (*N v The Department of Health in Northern Ireland*, 2003).

The grounds for the review were framed around the Human Rights Act (1998). In particular Article 2 concerning the applicant's right to life, Article 3 the right to freedom and harm from degrading treatment and Article 8 the right to respect for privacy for her family life.

On the day of the hearing in the High Court and immediately prior to the case being heard, the Department of Health in Northern Ireland agreed to fund the child's care package in full and all legal costs. The position adopted by the Department of Health not to fund a package of home care was likely to be untenable in view of the Human Rights Act and settling immediately before the hearing negated the payment of compensation to the child and her family.

The outcome of the judicial review was both of interest and concern to commissioners and managers of services. During exploratory fieldwork reported previously in Chapter 2, the view was frequently expressed by commissioners and managers that they had to find the money from somewhere to pay for increasing (and as yet unknown) numbers of ventilator-dependent children to live at home. The enactment of the Human Rights Act was considered to have provided families with a legal framework that could be used to force the NHS to pay for services as it was virtually impossible to argue that it was morally defensible to deny a child's right to a family life at home. Although children's community services have received substantial year-on-year increases in budgets, these increases have been insufficient to meet the growing demand for packages of community services to enable ventilator-dependent children to live at home.

6.2 What is already known about the costs of caring for children dependent on ventilators?

United Kingdom

A systematic search of the literature found 5 publications that estimated the costs of caring for ventilator-dependent children and young people in the UK (Figure 6.1). Only one study (Vick, 1996) was found to be a formal economic study, but was concerned with older adults on ventilators as opposed to children and young people. As adults access different services and funding streams it was not appropriate to use these costs as proxy estimates for children. The remaining 4 sources contained estimated costs concerning single hypothetical case scenarios and only included NHS costs concerning the ventilator and not the total support needs of the child (Jardine and Wallace, 1998; NHS Executive North West, 2000; North and South Thames PICU Network, 2001; Lenton *et al.* unpublished)

Although none of the cost estimates concerning children's support needs were derived from methodologically rigorous economic studies, all concluded that keeping a ventilator-dependent patient in hospital due to a lack of money or whilst contested decisions about funding were resolved, did not make economic sense. For example, Jardine and Wallis (1998) gave illustrative costs for home care, at 1997 prices, which varied from £240 per annum (with £500 capital costs) to £107,000 per annum (with £18,000 capital costs), depending on the child's medical condition and circumstances. Whilst the cost of an intensive care bed across studies varied from £1,000-£2,000 per day.

Europe

No economic evaluations were identified in English language journals.

Figure 6.1. Summary of cost estimates for adults and children on long-term ventilation in the UK.

Reference	Type of economic evaluation	Included costs	Excluded costs	Estimated costs of home care	Estimated costs of continued hospital stay	Price Year	Comments
Vick (1996)	Resource use of 11 ventilator-dependent adults	NHS/ Social services	Servicing of equipment and disposable supplies, informal carers Follow up hospital costs	Initial capital costs: Equipment £1,340 Staff £1080 Recurring costs 1 year: Staff £2,544- £82,521 (median £13,462)	£288 per day adult neurosurgery unit= £105,120 per year	1994/5 home care 1992/3 hospital	Adults require different service input than children
North and South Thames PICU network (2001)	Estimated costs of a typical child requiring 24 hour ventilation and home care	NHS	Social services, education, voluntary sector, parents. Follow up hospital costs, community and primary health services, ambulance and pharmacy.	Initial capital costs Equipment £14,330 Recurring costs 1 year: Servicing/maintenance £1,433 Disposables £8,656 Travel £16,740 Staff £127,389 Total recurring costs £154,218		1998	Not an economic evaluation
NHS NW Executive (2000)	Estimated costs of a typical child requiring 24 hour ventilation and care	NHS	Social services, education, voluntary sector, parents. Follow up hospital costs, community and primary health services, ambulance and pharmacy.	Initial capital costs: £20,000 Recurring costs 1 year: Consumables and servicing £ 7000 Staff £128,000 Total recurring costs £135,000	Paediatric intensive care unit: £1,300-£2,000 per day = £474,500-£730,000 per year	1998	Not an economic evaluation
Jardine et al 1998	Estimated costs of 3 hypothetical case scenarios	NHS	Social services, education, voluntary sector, parents, follow up hospital costs, community and primary health services, ambulance and pharmacy.	Initial capital costs: Range £500-18,000 Recurring costs 1 year: Consumables: Range £200-£7000 Service costs: Range £40-£400 Staff costs: Range £ 0- 100,000 Total recurring costs Range £240- £107,400		1997	Not an economic evaluation
Lenton et al (unpublished)	Cost analysis of a hypothetical child discharged from hospital. Child dependent on 24 hour ventilation and requiring 24 hour care	NHS	Social services, education, voluntary sector, parents Follow up hospital costs, community and primary health services, ambulance and pharmacy.	Set up costs: Equipment: £15,662 Recruitment and training: £14,872 Total £30,494 Recurring costs 1 year: Carers £127,389 Disposables £5,066 Travel £3,000 Maintenance £4,000 Management £7,000 Total recurring costs £148,017	Hospital 365 days @£1000 =£365,000	Not stated ? 2000	Not an economic evaluation

United States

The United States has a privately funded healthcare system, which makes the generalisability of economic studies to the UK problematic. Nonetheless, a systematic search of the literature was undertaken which revealed 5 publications with cost estimates (Figure 6.2). In addition, Aday *et al.* (1988) estimated the costs to insurance companies of home and hospital care for 141 ventilator-dependent children, but their published report was unobtainable as it was out of print. Almost all of these publications arose following two pieces of legislation. First, in many states, legislative bills had been passed to essentially eliminate 24 hour home nursing care in the belief that it was more costly than institutional care (Bach *et al.* 1992). This legislation was said to be responsible for over 55% of ventilator-dependent people having to live in acute hospitals or extended care facilities in Illinois. The main cost of home care was said to be trained nurses and state regulations prevented nursing activities from being carried out by cheaper unlicensed carers. As providing qualified nursing services at home was considered uneconomic, many hospitals had commissioned long-term ventilation units (so called extended care facilities) where many people on ventilators including children could be cared for by a small team of staff.

The second piece of legislation to influence practice was the Medicaid 'Katie Becket' Waiver. Medicaid was the major public funding source of medical services for adults and children with disabilities. Few Medicaid programmes reimbursed the purchase of equipment for use at home or home nursing care. President Reagan incorporated what became known as the Katie Becket Waiver into legislation after it became known that 5 year old Katie Becket who was dependent on a ventilator had been denied Medicaid funding for a package of care to live at home. Individual states were subsequently granted a pre-determined number of 'Waivers' to enable similar children to live at home (Beckett, 1996).

None of the publications summarised in Figure 6.2 were formal economic evaluations that met the criteria of a rigorous economic evaluation as described

by Drummond and Jefferson, (1996); but, as with the UK cost estimates, all concluded that care at home was cheaper (albeit to the insurance company) than care in hospital when the bulk of care was provided by parents and in situations whereby unlicensed carers were permitted to undertake nursing roles.

PAGE

NUMBERING

AS ORIGINAL

Figure 6.2. Summary of cost estimates for adults and children on long-term ventilation in the United States.

Reference	Type of economic evaluation	Included costs	Excluded costs	Estimated costs of home care per patient	Estimated costs of continued hospital stay per patient	Price Year	Comments
Hazlett (1989)	Chart review and analysis of Medicaid billing records of 15 children.	Health-service related	Non health related costs and parental input	31 days of home care \$2,844 - \$19,165	31 days of hospitalisation \$19,124 - \$52,526	1987-8	Not an economic evaluation. Only covers 1 month. Parents with insurance had to pay 10-20% of costs. Uninsured parents had to rely on state assistance and had lower levels of care.
Motwani and Herring (1988)	Survey of health service utilisation of 9 adults living at home. Hypothetical comparison of institutional care costs	Health service related	Non health service costs and carer input	Range of annual costs: \$50,240-\$61,970	Hypothetical annual cost: \$79,537	1986-7	Not an economic evaluation.
Fields et al (1991)	Hypothetical projections of service use with 6 children on Medicaid Waiver Programme. Hypothetical comparisons made.	Health service related	Non health service costs and parental input	Mean cost per year: \$109,836	Mean cost per year: \$188 909	1989-90	Not an economic evaluation. Hypothetical rather than actual service use described.
Bach et al (1992)	Telephone survey with 30 adults on Medicaid programme at home.	Health service related	Non health service costs and parental input	Mean cost per year: \$85,814	Mean cost per day automatically reimbursed for hospital care: \$648-\$718	1990-1	Not an economic evaluation
Sevick et al (1996)	Mailed questionnaire of resource use with 239 Medicaid supported adults.	Health service related	Non health service costs and carer input	Mean cost per month: \$7642		1994	Deals with adults only so findings not applicable to children.

The need for rigorous cost information

Review of the literature in Tables 6.1 and 6.2 revealed a substantial gap in the evidence base, which meant that policy makers had to make decisions without knowing the actual cost implications of managing a population of children dependent on ventilators. It was therefore decided to design and carry out an economic evaluation with the aim of producing accurate estimates of the support costs of ventilator-dependent children and young people.

Factors that influenced the design of empirical study

In Chapter 1, ventilator-dependent children were described as a heterogeneous group with only one thing in common - the ventilator. Each of the manifestations of ventilator-dependency has consequences for the provision of health and social care services, so the cost consequences of providing support could be considerable. The cost estimates that were summarised in Figure 6.1 revealed that most were simulated case studies looking at NHS costs concerning the ventilator as opposed to an entire package of care.

The cost burden and the costs of support are particularly difficult to estimate for a condition such as ventilator-dependency which may arise from, or result in, disabilities which cross traditional client group and service boundaries. In Chapter 2, findings from a study of hospital episode statistics failed to describe the in-patient resource use of this group of children as the appropriate ICD codes were not attributed as primary diagnoses by coders. Although further research is required before this approach is deemed unusable for calculating resource use, at the present time it is impossible to undertake 'top-down' cost of illness calculations, as the children dependent on ventilators cannot be distinguished separately.

It was decided that an economic evaluation conducted to rigorous standards was needed in order to address this information gap (Drummond *et al.* 1997). A full economic evaluation is defined as a comparative analysis of alternative courses of action in terms of both their costs and consequences (Drummond *et al.* 1997). Although desirable, it was not possible to complete a full economic

evaluation comparing the costs and consequences of children living in different locations (hospital, home, residential home), due primarily to time and resource constraints. In addition, as highlighted in Chapter 3, very little work had been undertaken with ventilator-dependent children to define or measure the outcomes or consequences of different locations of care. Finally, as described in Chapter 2, the known population of ventilator-dependent children in the UK is less than 150. Although this is likely to be an underestimate, it was decided that it would be unlikely that sufficient children could be recruited from different locations in order to compare the costs and consequences.

Because of the constraints identified above, it was decided to use a 'bottom-up' approach including a sample of children and young people to attach costs to the elements of their packages of care. The hypothetical estimates of costs described in Figure 6.1 were dominated by a health service perspective and rarely included the perspectives of other agencies such as social services, education authorities or the voluntary sector. None of the cost estimates included the input of parents or the financial impact on the family, even though a number of narrative articles and actuarial assessments acknowledge that the improved health outcomes associated with home care and lower health service costs have been achieved at the cost of parents (for example, Leonard *et al.* 1993; Carers UK, 2002). In order to fill this significant gap in the evidence base, it was decided initially to undertake a study of costs and consequences from a societal perspective that aimed to measure the cost burden to the NHS, social services, education authorities, voluntary sector and parents.

6.3 Quantitative study to estimate the costs of caring for ventilator-dependent children

Methods

The process of costing used in a 'bottom-up' or client centred approach has 3 connected parts:

1. The collection of service receipt or utilisation, and equipment data from the client;
2. The costing or pricing of each of the services or equipment used, and
3. The combination of these 2 sets of information in order to cost full packages of care (Beecham, 1995).

The collection of service receipt or utilisation and equipment data

It was decided to adapt a validated data collection tool called the Client Service Receipt Inventory (CSRI) as this had been used previously to assess service use and cost consequences of similar client groups. One such example, was a study by Beecham *et al.* to assess the service and cost consequences of supporting a sample of young people with hemiplegic cerebral palsy (Beecham *et al.* 2001).

Theories underpinning the CSRI

Economic theory

The CSRI bases cost measures on long-run marginal opportunity cost. Since national policy intentions are to provide long-term community services for ventilator-dependent children so that they can be discharged from hospital, it would not be credible to measure only short-run cost implications. Marginal cost reflects the addition to total cost attributable to the inclusion of one more child (Torgerson and Spencer 1996). Opportunity costs reflect the resource implications of opportunities forgone rather than amounts spent (Palmer and Raftery, 1999). The opportunity cost measures the true private or social value of a resource or service, based on its value in the best alternative use (Beecham, 1995). For example, measuring opportunity and marginal costs will be especially important in relation to the appropriate use of intensive care beds. In theory, the use of long-run marginal costs allows examination of the difference between providing in-patient care versus community-based home care and the potential impact of either option to available budgets. According to Beecham (1995), short-run marginal costs are inappropriate for most costing tasks as they only include revenue costs and do not take account of the full costs of creating new services. However, knowledge about the present time is said to be more certain than knowledge of the future, so the convention is to

use short-run average costs which include both revenue and capital elements as an approximation for long-run marginal costs.

This method encompasses the resource implications of all elements of a service or care package and is underpinned by 4 basic cost rules:

1. Costs are measured comprehensively at an individual client level and range over as many service components of care programmes or 'packages' as is relevant;
2. Variations between clients, facilities or areas of the country are taken into account;
3. Comparisons are made on a like -for-like basis, and
4. The final guiding principle underpinning this method is timeliness. The year chosen to calculate the costs data should be as up to date as possible to enhance the validity and utility of the results. Beecham recommends that service costs information should apply to the time in which the policy is to be implemented or the service used.

Model of disability

The theoretical perspective adopted by this method of economic evaluation is aligned with the medical model of disability as costs are compared with service use from 'normative' populations, although it could be argued that this particular economic evaluation is also underpinned by elements derived from the social model of disability, as the evaluation aims to take a social perspective.

Adaptation of the CSRI

The CSRI had been used widely in previous economic studies and examples of the questionnaires were obtained for reference purposes. The CSRI contained 5 core sections (listed below) that formed a framework within which responses to questions were structured to facilitate cost estimation. Using findings derived from the qualitative exploratory study undertaken with 18 children and their families (described previously in Chapter 4), each of the 5 sections were

adapted to reflect the range of equipment, services and options available to children dependent on ventilators:

Sociodemographic survey

The opening section covered background and client information (such as date of birth, sex, ethnic grouping and method of child's communication). In addition, there was space to record the child's diagnoses and as many of these children had long histories of accessing hospital services, an estimate of the total length of time that the child spent in hospital during their life was recorded.

Current accommodation and use of transportation

The second section concentrated on describing the child's living situation. Where the child lives is usually a major component in terms of the costs of care. The whereabouts of the child was recorded over the previous 12 months (hospital, home, residential) and any movements between accommodation types were noted. The nature of the family accommodation was recorded, including the tenure, a simple description of the size of the unit, number of other residents and, as previous work had indicated that many homes required expensive adaptations, a survey of adaptations was included. As it was anticipated that a number of children would be in the care of social services, this section also documented the status of the child, whether they were 'looked after', and the level of contact with their families. Finally, as many disabled children were known to have problems with accessing affordable and accessible transport, this section documented all the methods of transport used by the child (excluding ambulance and school transport which were recorded in other sections).

Education and training profile (of the child and parents)

Section 3 recorded the nature, type and frequency of education or training that school age children and young people received and how they were transported to educational facilities. As many of these children have statements of educational need, their status was recorded and any additional support services provided to meet additional needs was documented. As many children also

received additional health services whilst at school, a section was included to document service receipt received.

In order to understand the level of educational qualifications that children were aiming for, examination courses and qualifications gained were documented together with the child's aspirations or plans for the future. It was also decided to document maternal and paternal educational and professional qualifications so that comparisons could be made with national census data.

Employment and income (for children over 16, and parents)

Research has shown that families with disabled children have a reduced capacity to earn a living because of their caring responsibilities and there is a higher incidence of family breakdown leading to poverty. Section 4 concentrated on collecting information on employment history, income and household debt of parents and if appropriate young people who used ventilators. This section also recorded receipt of social security benefits (although in strict economic terms these should be considered as transfer payments that do not represent an aggregate cost to society). Data on changes in benefit over the past 12 months were also recorded, as many children were known to move between hospital and home settings thereby triggering access to different benefits.

Services Receipt

The services receipt is the core of the CSRI. Services for children who are dependent on ventilators are known to be delivered and received in a fragmented way with many agencies and organisations providing a variety of services and equipment. This section identified all the health, social and voluntary services, and equipment received as an in-patient, out-patient, at home or at a community facility (including when purchased, type of service, name of provider, frequency and mode of attendance or contact). As many families may also receive unpaid services, this section detailed the input of informal carers in terms of the time spent (frequency and duration of support) and the tasks undertaken.

For services likely to absorb a high proportion of care package costs, more precise detail was collected on the type of hospital and ward, and the exact length of each in-patient admission and why the child was admitted.

Finally, as exploratory fieldwork had shown that parents frequently accessed support over the telephone and their children used complementary therapies, questions were included to record the amount and frequency of telephone contact with services, use of the internet and complementary therapies. For teenage children, information was documented about the arrangements for transfer to adult services.

Satisfaction with services

Questions about satisfaction with services and the gaps in service availability were included throughout the questionnaire. According to Beecham (1995) neither is needed for cost calculation, but both offer useful insights when making judgements about the cost of a child's care package and the fulfilment (or not) of the child's needs.

Satisfaction levels have been used widely as an indicator of the quality of services provided (Aspinal *et al.* 2003). On a similar vein, Locker and Dunt (1978) suggested in their review of methodological and theoretical issues around satisfaction of care, that particularly in long-term care:

'Quality of care can become synonymous with quality of life, and satisfaction with care can be an important component of life satisfaction.'

There is a substantial literature around theoretical and methodological issues and the strengths and weaknesses of using satisfaction with services as an outcome measure for evaluating interventions. To do justice to this large literature is beyond the scope of this thesis. For a review of some of the theoretical and methodological issues, reference can be made to Ruggeri (1994) and Aspinal *et al.* (2003).

The CSRI assesses very broadly the quality of contact with specific services and overall quality of the management of care packages on a 5-point Likert scale: completely satisfied, mostly satisfied, mixed, mostly dissatisfied, or completely dissatisfied. Questions are designed to measure the extent to which services received gratified the child's or parent's wants, wishes or desires about a service or intervention, including: the felt adequacy of the service provided; the appropriateness of the child's living conditions and surroundings, and the availability and accessibility of transport. Ordinal questions were included to ascertain if services were being reviewed on an annual basis and whether there were any logistical issues about the delivery of services in the community.

Service availability was assessed additionally by whether children needed access to:- more of, the same, or less of a particular service. Finally, questions ascertained whether a child's needs had been met or not by the services provided and whether the child had any needs that required addressing, and whether they had the appropriate aids, adaptations and equipment.

A copy of the adapted CSRI tool can be found in Appendix 3.

Piloting of tool

The CSRI was piloted with 2 mothers in order to ensure that the language and presentation was clear, the data required was realistic, and to provide an indication of the time required to complete the questionnaire. Piloting revealed that the style and content of the questionnaire was received positively and only a few minor changes were made to language and phraseology.

Confirmation of time horizon of study

The CSRI collects retrospective data on service utilisation and service-related issues. The retrospective period prior to the date of the interview is a compromise between the accuracy that comes from not asking respondents to cast their minds back too far and the comprehensiveness which can only come from allowing sufficient time to elapse for some uncommon but potentially

expensive services to be used (Beecham and Knapp, 1992). Piloting confirmed that parents could recall:

- Health, social, education and voluntary service utilisation over the previous 12 months;
- Low volume disposable supplies over the previous 6 months, and
- High volume disposable supplies over the previous 1 month.

It was therefore decided to collect service utilisation over the previous 12 months and where necessary, resource utilisation was multiplied to give resource use for the previous 12 months.

Potential limitations to data collection

Piloting also identified 4 potential limitations to data collection:

(i) The length of time required to complete the questionnaire

The original CSRI was 10 pages long and took around 40 minutes to complete. The 26 page CSRI used in this study reflected the huge range of resources used by this group of children. Piloting showed that the questionnaire would take up to 2 hours to complete and in addition was accompanied by another questionnaire to measure health-related quality of life and a qualitative semi-structured interview. In total, it was estimated that families would need to give up a minimum of 3.5 hours of their time.

(ii) Access to financial information (especially amongst minority ethnic women)

The study aimed to recruit a subgroup of families from minority ethnic backgrounds and the data collection technique relied on the memory recall of parents or their ability to access domestic financial records. One of the pilot mothers who was of Asian origin said that most Asian women did not have knowledge of the family finances and she did not think that the men from her community would see it as their business to discuss issues concerning the disabled child with a researcher. Both mothers in the pilot had a detailed knowledge of their child's service and resource use, but neither had either kept

or had access to records of the costs concerning the adaptations to their homes or the complex arrangements concerning the means tested disabled facilities grant that were used to fund their adaptations.

(iii) Complexity of funding for domestic transport

It became clear that issues around the funding of domestic transport were complex and there was a danger of double counting. Both pilot families were in receipt of welfare benefits that were provided specifically to help their child access domestic transport. One family had obtained a means tested grant via the Motability scheme to purchase a car, but could not remember the specific financial details concerning their own financial contribution or whether this was covered by the child's mobility component of the disabled living allowance.

(iv) Complexity of calculating opportunity costs around lost employment of parents

At the conception of the study, the aim was to collect data from a societal perspective, but piloting revealed how difficult it was likely to be to calculate the opportunity costs of lost or reduced employment for unpaid carers (principally mothers). For example, the Asian mother said that her role within the family was that of wife and mother, but if she did not have a disabled child she would be expected to contribute to the family business for which she was unlikely to be paid wages. The other mother had her own business and worked prior to the birth of her disabled child. She was currently receiving incapacity benefit as well as caring for her child, and was also pregnant again and about to claim statutory maternity benefits. The grandmother took a substantial role in providing support for her daughter, but was said to see this as her natural role. She managed her daughter's business whenever required for which she was paid a share of the profits but was officially retired and received both a state and occupational pension.

Impact on study perspective

With additional written consent, it may have been possible to obtain information from the relevant housing authority or Motability scheme, but it was decided that there would be insufficient time to do this. Beresford and Oldham (2002) also encountered similar problems concerning the availability of accurate financial

data when researching the housing needs of disabled children and their families. They found that local authorities did not keep information on the financial contribution of parents towards disabled facilities grants. Given the time constraints and difficulties identified concerning access to specific costing information required to complete an economic study from a societal perspective, it was decided to prioritise the time available to costing detailed formal health, social, education and voluntary sector service and resource use, and exclude the costs of domestic transport, housing adaptations, and informal care from the total annual support costs for the entire group of children. If individual parents were able to provide accurate data then it was decided to present illustrative costs of individual families.

Aim of study

The aim of the study was to provide an estimate of the long-run marginal service costs of supporting a stratified sample of 35 children and young adults who were dependent on ventilators in hospital, home and residential settings over 12 months.

It was decided to focus the study on 5 areas of interest which are reflected in the following primary and secondary research questions:

Primary Research question:

1. What are the long-run marginal service costs of supporting ventilator-dependent children in hospital, home and residential settings over 1 year?

Secondary research questions:

2. Can costs be compared to 'normative' and other groups of children to give an order of magnitude estimate of the 'excess' costs?
3. Can comparisons be made between the costs of those children living in hospital and those living at home?

4. Can the case study data be used to build up examples of costed packages of care?
5. How satisfied are parents and children with the services they receive?

Perspective of study

The study was undertaken from the perspective of the NHS, social services (excluding the cost of housing adaptations), education services and the voluntary sector.

Time Horizon

Long-run marginal costs were calculated for the previous 12 months.

Sample

As described previously in Chapter 4, it was planned to build up case studies around 35 children and young people who were ventilator-dependent and aged from 1 year to 19 years. The children and their families were recruited via 11 hospital consultants in the UK. A theoretical sampling strategy was used in order to minimally include the following in the final sample and among those interviewed:

- 10 children age 5 years and under;
- 10 children age 6-12 years;
- 10 children and 13-19 years;
- Representation from both boys and girls in each age band;
- Representation from children who required assisted ventilation for 24 hours per day and children who used it for less than 24 hours per day in each age band;
- A mix of children who lived in hospitals, domestic settings and 'looked after' children in the care of social services in each age band, and
- Representation of parent(s)' views.

Recruitment was successful and all of the theoretical requirements of the sample were satisfied apart from including a 'looked after' child living away from their family in each age group. The barriers to accessing 'looked after' children as interviewees have been explored previously in Chapter 4. The resultant sample shown in Figure 6.3 included 35 ventilator-dependent children (12 girls and 23 boys); 12 used ventilation 24 hours per day, 7 were living in hospital at the time of interview with the remainder living at home. Of those living at home 4 had been transferred from hospital within the previous 12 months. Interviews were conducted with 19 children and parent(s) or those acting in loco parentis of all 35 cases. Eight families were from ethnic minorities and one child was 'looked after' by social services. Nine families described themselves as a 'lone' parent family.

Figure 6.3. The sample.

Age range	No	Sex	Diagnosis	Type of ventilation	Location of child	Lone parent	Ethnic minority	Looked after	Child interview	Parent interview
5 years and under	1	F	Spinal injury	PPV tracheostomy (24 hours)	hospital	✓	✓			✓
	2	M	Congenital	PPV tracheostomy (24 hours)	home					✓
	3	M	Congenital	PPV (when asleep)	home	✓	✓			✓
	4	F	Congenital	PPV (when asleep)	home	✓	✓			✓
	5	M	Congenital	PPV tracheostomy (20 hours day)	hospital					✓
	6	F	Congenital	PPV mask (night time)	home				✓	✓
	7	M	Congenital	PPV tracheostomy (24 hours)	home					✓
	8	F	Congenital	PPV tracheostomy (when asleep)	home		✓			✓
	9	M	Congenital	PPV tracheostomy (24 hours)	home	✓				✓
	10	M	Spinal/head injury	PPV tracheostomy/PPD (24 hours)	hospital				✓	✓
	11	M	Congenital	PPV tracheostomy (20 hours day)	hospital		✓			✓
6 – 12 yrs	12	M	Congenital	PPV mask (Night time)	home	✓			✓	✓
	13	M	Congenital	PPV mask (when asleep)	home					✓
	14	M	Congenital	PPV tracheostomy (24 hours)	home					✓
	15	F	Spinal/brain injury	PPV mask (when asleep)	home				✓	✓
	16	M	Congenital	PPV tracheostomy (when asleep)	home	✓				✓
	17	M	Congenital	PPV mask (when asleep)	home				✓	✓
	18	M	Congenital	PPV mask (when asleep)	home	✓				✓
	19	M	Spinal/brain injury	PPV tracheostomy (24 hours)	hospital			✓		✓
	20	F	Congenital	PPV mask (when asleep)	home				✓	✓
	21	F	Spinal/brain injury	PPV tracheostomy (24 hours)	hospital				✓	✓
	22	M	Congenital	PPV mask (night time)	home		✓		✓	✓
	23	M	Spinal/brain injury	PPV tracheostomy/PPD (24 hours)	home				✓	✓
	24	M	Congenital	PPV tracheostomy (24 hours)	home		✓		✓	✓
	13 – 19 yrs	25	M	Congenital	PPV mask (night time)	home				✓
26		F	Congenital	PPV mask (night time)	home				✓	✓
27		F	Congenital	PPV mask (when asleep)	home	✓				✓
28		F	Congenital	PPV mask (night time)	home					✓
29		M	Congenital	PPV nasal pillows (night time)	home	✓			✓	✓
30		M	Congenital	PPV mask (night time)	home				✓	✓
31		M	Spinal/brain injury	PPV tracheostomy (24 hours)	home		✓		✓	✓
32		F	Congenital	PPV mask (night time)	home				✓	✓
33		M	Congenital	PPV mask (night time)	home				✓	✓
34		M	Spinal/brain injury	PPV (tracheostomy) 24 hours	hospital				✓	✓
35		F	Spinal/brain injury	PPV mask (night time)	home				✓	✓

Key: PPV: positive pressure ventilation PPD: Phrenic pacing device

Ethics

Ethical issues and consent procedures were as described previously in Chapter 3.

Data collection

Data were collected in a face-to-face interview with parent(s) and where appropriate the ventilator-dependent child between September 2001 until March

2003. Interviews were conducted at the family home or a quiet room at the hospital. As many of the children's packages of care were complex, interviews were also tape recorded so that reference could be made at a later date to check that information recorded on the questionnaire was correct. A series of prompt cards were used to aid the completion of the questionnaire. These laminated cards contained indicative lists of accommodation types, different national and local services, social security benefits, equipment and supplies etc. Parents and children could scan these cards at appropriate times during the interview to ensure that they described all of the resources used and services they had contact with. Some parents also provided comprehensive stock lists and monthly printouts detailing exact resource use and costs.

The costing or pricing of each of the services or items used

For each of the services used by the children and young people a unit cost per hour of service was estimated which was a close approximation to the long-run marginal opportunity cost. The Personal Social Services Research Unit produces an annual compilation of unit costs for health and social care (Netten and Curtis, 2002). Netten and Curtis use a validated methodology to estimate a unit cost per hour of service (Beecham, 1995). This method requires a clear description of service inputs to which a financial valuation is attributed to each input, then the measurement of service activity or output over time is computed to produce a unit cost per hour of service. Factors such as capital costs, salary costs, salary on-costs, past and current training costs, and capital overheads are included in the unit cost calculation.

Costs were attributed at 2002 prices, which were the latest available. Cost calculations published prior to 2002, were brought up to 2002 prices by using inflation indices provided by Netten and Curtis (2002). Prices were calculated excluding London cost of living allowances unless indicated.

The annual compilation of nationally applicable unit costs (Netten and Curtis, 2002) was the main source of valuation used. Unit costs were adjusted in the light of the frequency and duration with which each child used each service, allowing calculation of the individual support costs in the 12 months prior to

interview. In addition, 6 other sources of valuation were used to calculate the costs of resources not covered by Netten and Curtis (2002), including:

- National data on pay, working conditions and welfare benefits available on Government websites;
- NHS Finance Manual (Department of Health, 2002a);
- British National Formulary (BNF, March 2002);
- Family sources;
- Market prices, and
- Previous economic evaluation of oxygen concentrator versus oxygen cylinder use (Heaney *et al.* 1999).

In relation to equipment, it was assumed that the device would have a life of 5 years and would cost 15% of the total cost per year to service and maintain. Exceptions to this rule were items such as wheelchairs, which on average last children up to 3 years before they outgrow them.

As stated previously, the costs of informal care or lost employment can be high for families with disabled children, but due to the complexity of the task and limited time allowed they have not been estimated in this study. Likewise, it was not possible, due to lack of available information, to estimate the cost of housing adaptations or transport across the entire group. However, one parent could provide detailed information, so although costs are not included for the entire group, a description of resource use and individual costs are given as examples when available.

Data analysis: The combination of resource use with costing and pricing data in order to cost full packages of care

Data derived from individual case studies were entered into an Access™ database (Microsoft, 2000) and converted into Excel™ (Microsoft, 2000) and SPSS™ version 11 for analysis (SPSS, 2001). Costs were ascribed from the different sources of valuation outlined above.

For the purposes of analysis costs were calculated for:

- The entire group of 35 children and young people;
- 7 children living in hospital during the previous 12 months;
- 24 children living at home during the previous 12 months;
- 4 children who were discharged from hospital to home during the previous 12 months, and
- Individual children.

To compare characteristics and service use of children and young people dependent on ventilators with the general population, 4 data sources were used. *The Living in Britain 2001 Survey* (Office National Statistics (ONS), 2001) contains information from the General Household Survey, which has documented major changes in British households, families and society since 1971. *The British Household Panel Survey (BHPS)* (ONS, 2002) is a multi-purpose study that has been following the same representative panel of individuals since 1971. *The 1970 Birth Cohort Study* originated in the British Birth Survey of over 17,000 babies born in Britain in one week in 1970 (ONS, 2002). The survey has followed the health, education, social and economic circumstances of cohort members. Finally, reference was made to *UK Labour Force surveys* of 2002 to ascertain baseline employment data (ONS, 2002).

In addition, findings were compared with a study using the same methodology with a group of young adults with hemiplegic cerebral palsy living at home (Beecham *et al.* 2001). As it was not possible to access children living in residential placements, findings were also compared with support costs of severely disabled young people living in 2 residential settings (Netten and Curtis, 2002).

Results

Accommodation

At the time of interview, 24 children had been primarily living at home with their families over the past 12 months, 7 children were living in hospital and 4 children were currently living at home having been discharged from hospital in the previous 12 months.

It emerged that 43% of children (15) had experienced a change in their accommodation (some on more than one occasion) and had lived somewhere else for more than 1 month in the last 12 months, including: from home to hospital (6); hospital to home (4) neonatal intensive care unit (1); paediatric intensive care unit (6); children's ward (1); adult intensive care unit (2) and adult spinal injuries unit (1). Six children had lived in hospital for between 12-28 months, one child had lived in hospital for more than 5 years.

Satisfaction with child's current living situation

A mixed picture emerged concerning the appropriateness of where children were living. Twenty-six parents (74%) were either completely satisfied or satisfied that their child was living in the most appropriate place and all of these children lived primarily at home. Four parents (11%) had mixed feelings, and 5 (14%) were either dissatisfied or completely dissatisfied; the latter being parents of children in hospital.

Housing

A higher percentage of families with ventilator-dependent children (13/34, 38%) rented their homes from local authorities or housing associations than the general population (18%*) or families with a child with hemiplegic cerebral palsy (22%). Fewer families (19/34, 56%) lived in an owner occupied house compared with the general population (68%*) or families with a child with hemiplegic cerebral palsy (59%). * General Household Survey Living in Britain 2001

Adaptations to housing

Over 85% of families (30/35) had undertaken or were currently undertaking work to adapt their domestic homes to accommodate the needs of their disabled child. Of the sample:

- 4 lived in houses with no adaptations;
- 1 lived in a bungalow with no adaptations;
- 18 lived in houses with adaptations;
- 2 lived in bungalows with adaptations;
- 5 lived in purpose built dwellings, and
- 5 were undertaking work on their houses so that their child could be discharged.

Family homes ranged in bedroom number from 2 to 6 bedrooms, (15 families had 4 or more bedrooms, 17 families had 3 bedrooms, and 2 families had 2 bedrooms).

The number of adults over age 18 ranged from 1-3 (one adult (7), 2 adults (22), 3 adults (5)). Mean family unit size was 3.8 persons (range 3-7). Families had on average 1.9 children (range 1-5), which is slightly higher than national trends (1.65 *). * General Household Survey Living in Britain 2001

As stated previously, it was not possible to obtain detailed costings concerning accommodation and housing adaptations for the entire group of children. Twenty-nine families had received or were applying for a means tested benefit of up to £20,000, called the disabled facilities grant, in order to adapt their homes. Almost all parents who owned their homes had increased their mortgages or remortgaged their house to pay for extensions and building work. One family had also moved into a state of the art architect designed house within the previous 12 months. One local authority was purchasing a house on the domestic housing market for £120,000 and was then intending to spend £38,000 on adapting the accommodation.

It was possible to undertake a survey of adaptations that families had undertaken to their homes (Table 6.1). The survey revealed that extensive work had been undertaken, including adapting outside spaces, providing access to the house, major extensions and remodelling, and installing new fixtures and fittings. In addition, 23 families reported that they needed to undertake additional work to their homes (such as bigger bathrooms) to meet the changing needs of their disabled child.

Table 6.1. Survey of adaptations to 30 domestic homes.

Drive	Garage	Garden	Front door/backdoor	Entrance hall
5 - alterations to drive including ramps/tarmac/widening/safety gates	5 - converted garage into accommodation to disabled child	6 - had adapted garden including tarmac/ramps/paths	12 - had alterations to front door including widening/ramps/canopy	5 - had alterations to hall including widening/ramps/widening doors
Family living space	Storage	Extension	Disabled child's bedroom	Disabled child's bathroom/shower room
12 - adapted the family living space including widening doorways/ceiling hoist/flooring/piped oxygen	12 - additional storage space added	9 - extensive building work to provide extra accommodation for disabled child and carers	13 - adapted child's bedroom including ceiling hoists/wide doorways etc	13 - adapted bathroom including wide doorways/ceiling hoists/fixtures and fittings/ adapted toilet and seating

Illustrative costs of undertaking adaptations for one family

One parent was able to provide detailed information concerning the aids installed and adaptations undertaken to her house with the help of a means tested disabled facilities grant.

The work was carried out in order to provide an additional bedroom and an adapted bathroom that were accessible to a 15 year old boy. In addition, the family wanted extra room (lounge and kitchen) to accommodate their son's carers and storage for his equipment (not funded by the disabled facilities grant). A disabled facilities grant of £20,000 was awarded by the local authority in 2001 and his parents re-mortgaged their house to pay for their £25,000 contribution. The alterations would, if necessary, provide suitable accommodation for the rest of the boy's life, but he had plans of his own to live independently as an adult.

As the boy had a degenerative condition and his life expectancy was unknown the costs per year of the disabled facilities grant were calculated assuming that he would move into an independent living scheme in 3 years time at 18 years old. The long-run marginal cost would therefore be £6,666 per year.

The cost to the parents was difficult to calculate as their mortgage was spread over 25 years and will be subject to varying rates of interest. In addition, the market value of their house had increased substantially by the addition of a 2-storey extension providing 4 extra rooms. This accommodation had also been designed so that it could be self-contained, by the closure of an adjoining door. The mother said that if her son moved into independent accommodation then they would consider renting their self-contained annex at a commercial rate.

Beresford and Oldham (2002) undertook a survey of the housing needs of disabled children in 2000. Their report provides estimates for the average cost per disabled facilities grant awarded to families of disabled children in one local authority (£15,750, 2000 prices). Given that the maximum disabled facilities grant is set at £20,000 unless there are exceptional circumstances, and the adaptations are meant to ensure that the child's housing is suitable until adulthood, this figure would not make a significant difference to the total annual support costs of ventilator-dependent children.

Satisfaction with domestic accommodation

A mixed picture emerged concerning the domestic homes where children were living or would live on their discharge from hospital. Seventeen parents (48%) were either completely satisfied or satisfied with their domestic housing. These parents tended to live in either public or private sector homes that had either been purpose built for their child or had been extensively adapted to meet their child's needs. Six parents had mixed feelings (18%), and 10 (29%) were either dissatisfied or completely dissatisfied; the latter being parents whose homes either were wholly or partly inappropriate for a disabled child to live in. As described previously in Chapter 4, inappropriate housing also had a major negative impact on the quality of life of the child, their family and working conditions of their employed care teams.

Domestic transportation

Of the 34 families surveyed, 7 (20%) did not own a vehicle whereas in the general population slightly more people (28%*) did not own a vehicle. One young person had his own car and was learning to drive; 17 families had a saloon car; and 9 families had a people carrier/or a van plus adaptations.

* General Household Living in Britain Survey 2001

The 35 children also used a variety of other methods of transportation, including: black cabs (4); minibus (8); dial a ride/ nexus bus service (2); metro train (1); bus (5); relatives' cars (5); hospital minibus (3); campervan (1); nurses' own car (1); and hospital taxi (1).

It was not possible to obtain detailed costings concerning transportation for the entire group of children. Most parents had obtained a van or people carrier through a Motability scheme, which is a not-for-profit organisation that provides mobility solutions for disabled people. Some families had been awarded additional money by Motability to undertake expensive adaptations to vehicles such as in-vehicle hoists, slings and hydraulic ramps. The financing of the Motability scheme was complex and most parents did not have detailed information to hand. Twenty-four children also received the mobility component of the disability living allowance, which was designed to pay for the child's transport needs so there was a danger of double counting. Many families had more than one car that was used by their disabled child and parents had used a range of financial products to purchase second vehicles.

Illustrative costs of buying an adapted van for one family

One parent was able to provide detailed costings of the adapted van that the family had purchased independently. The van was adapted with a lift and special seating and the total cost to the family was £28,000 in 2002. They expected the vehicle to be replaced in 5 years, which would produce a long-run marginal opportunity cost of £5,600 a year (excluding interest payments on the bank loan and trade-in value). Their child also received the mobility component of the disability living allowance at the higher rate of £37.40 per week.

Satisfaction with the availability of transport

The availability of accessible and affordable transport varied across the sample. Sixteen respondents (46%) were either completely satisfied or satisfied and these were families who had adapted or accessible transport (such as vans or people carriers), or they lived in areas where public transport was accessible to disabled people (such as modern tram and metro systems). Six respondents (17%) had mixed feelings, and 13 (37%) were either dissatisfied or completely dissatisfied. The latter were either parents of children in hospital who were isolated socially by the lack of transport available to them, or parents and young people who did not have ready access to transport so that they could go out whenever they wanted to.

Employment and income of families

Nine families (9/34, 26%) described themselves as lone parent families and in all cases the lone parent was the mother. Fifteen out of 34 families (44%) had no adult over 16 years in paid employment, which is higher than the national average of 5%[¶]. Eleven mothers (32%) had paid employment; the remainder described themselves as housewives or carers, and 2 did unpaid voluntary work. Of the 25 fathers on whom information was provided, 18 (72%) were employed, 2 were unemployed (8%) and the remainder described themselves as carers (3) or other (2). [¶] Labour Force Survey 2002.

Seventy-six percent of families had a total household income of under £25,000 per year, which in 2001 was classified as the mean total household income in the UK (Table 6.2). Lone mothers mainly occupied the lower income brackets and most said that they did not receive child support payments from fathers. This level of income is similar to families with a dependent child in the Living in Britain survey of 2001, where although the income bands are slightly different, similar trends are evident, with lone mothers experiencing most financial hardship (Table 6.2).

Table 6.2. Total Household Income.

35 case studies with ventilator-dependent children		Living in Britain 2001 Survey	
Yearly income	Ventilator-dependent children	Yearly income	Families with dependent children
£0 - £10,000 year	5/34 (15%)	£0 - £15,600	19% married couple 72% lone mothers
£10,001 - £15,000 year	9/34 (26%)		
£15,001 - £20,000 year	7/34 (20%)	£15,601 - £26,000	20% married couples 19% lone mothers
£20,001 - £25,000	5/34 (15%)		
25,001 - £30,000 year	4/34 (12%)	Over £26,000	61% married couples 9% lone mothers
£30,001 or more per year	4/34 (12%)		

Household debt

Fifty-six percent of families had significant debts, excluding mortgages. Of this group, 16 families had debts of up to £10,000 and 3 families had debts of between £15-20,000, the latter being accounted for by vehicle loans to purchase accessible transport. This picture is not dissimilar to national norms of debt in 2002 (McMahon, 2003), although for these families the debt was accumulated in most part by buying essential rather than unessential or luxury goods.

Receipt of welfare benefits

Thirty-four out of 35 children and their families said they had received social security benefits (the exception was the child 'looked after' by social services). The range of benefits received is outlined in Table 6.3.

For the 34 children and families who received benefits, the average yearly payment was £7,380 (SD £3,395); payments ranged from £821 - £15,334. The total transfer payment for all 34 children was £258,318 over the previous 12 months. Children and their families had access to different benefit streams depending on whether they were living in hospital or at home. For example, the disability living and mobility allowance could not be claimed until the child had been discharged from hospital. Whereas 7 parents were able to claim reimbursement of their travel costs to and from hospital, which for one parent with a child living in hospital amounted to £3,000 over the previous 12 months. Parents appeared to be claiming all the disability-related benefits on behalf of their children but it was difficult to assess without closer scrutiny of individual

finances whether they were claiming their full entitlement to council tax and housing benefit.

Table 6.3. Welfare benefits received by families.

Welfare benefits received by families	
Child benefit	34/34
Invalid care allowance / Carers allowance	14/34
Council tax benefit	12/34
Disability living allowance	Higher 26 Medium 2 Lower 1
Disability living allowance (mobility component)	Higher 8 Medium 16
Housing benefit	6 families
Incapacity benefit	10 parents
Transport costs to and from hospital	7
Income support	9
Disability premium	4
Severe disability premium	1

Highest maternal and paternal qualifications

Maternal

Seven out of 34 (20%) mothers had no qualifications; 6/34 (18%) achieved one or more general certificates in education; 17 (50%) attained vocational qualifications such as hairdressing or nursing and 4 (12%) were awarded a diploma or degree.

Paternal

Thirteen out of 33 (39%) fathers had no qualifications; 7 (21%) achieved vocational qualifications and 13 (39%) achieved a diploma or degree.

In comparison, adults in the 1970 British Cohort Study when followed up at age 30 years old, around 90% had at least one certificate of education, general certificate of education or a Scottish School Certificate and just over 4% of people had gained a diploma, degree or higher degree. No information is provided concerning vocational qualifications.

In the current study, families with a parent with either a diploma or degree fell into income brackets above the national average wage and account for almost all of the 24% of families whose income was over £25,000 per year (Table 6.2).

Service use of 24 ventilator-dependent children living at home in the previous 12 months

Service use was calculated for ventilator-dependent children living at home during the previous 12 months and compared with population surveys and young people with hemiplegic cerebral palsy (Table 6.4).

Table 6.4. Service use of 24 ventilator-dependent children living at home in the previous year.

Service use of 24 ventilator-dependent children and young people living at home (Use per year unless otherwise indicated)	% of people using service						Normative data children 5-15 years		Hemiplegic Cerebral Palsy (n=81) age 18-24 years		Normative data Young adults age 16-44 ¹ Young adults age 18-25 ²	
	Ventilator-dependent children aged 1-20 years (n=24)		Normative children 0-4 years		Normative children 5-15 years		Hemiplegic Cerebral Palsy (n=81) age 18-24 years		Normative data Young adults age 16-44 ¹ Young adults age 18-25 ²		Normative data Young adults age 16-44 ¹ Young adults age 18-25 ²	
	% (n)	Mean number contacts	Range	%	Mean number contacts	%	Mean number contacts	% (n)	Mean number contacts	%	Mean number contacts	
Inpatient stay in previous 12 months	75% (18/24)	1.8	1-4	9% ¹	7 nights ¹	4% ¹	4 nights ¹	12% (10)	7% ¹	4 nights ¹		
Average length of inpatient stay		9.7 nights	1-63									
Outpatient / A&E attendance in last 3 months	100% (24/24)			13% ¹		9% ¹		38% (31)	12% ¹			
General practitioner (face to face)	62% (15/24)	7.7	1-25					74% (60)	72% ¹	4 ¹		
GP repeat prescriptions	87% (21/24)	12										
Health visitor	100% (7/7)	6.2	1-12									
Practice Nurse	16% (4/24)	1										
School doctor	85% (11/13)	1										
School nurse	100% (13/13)	82	1-190									
Community Paediatrician	8% (2/24)	2										
Children's community/district nurse	45% (11/24)	16.5	2-52									
Dentist	96% (23/24)	2.1	1-6					32% (26)	55% ²			
Orthodontist	12% (3/24)	2.3	2-3									
Optician	8% (2/24)	1.5	1-2					2% (2)	27% ²			
Specialist doctor/surgeon	96% (23/24)	5.5	1-11					35% (28)	16% ²			
Orthotist	12% (3/24)	2.3	1-3					4% (3)				
Chiroprapist	8% (2/24)	3	2-4					5% (4)	2% ²			
Physiotherapist	62% (15/24)	57.4	2-190					12% (10)	4% ²			
Speech or occupational therapist	50% (12/24)	14.2	2-48					5% (4)	1% ²			
Dietician	37% (9/24)	2.3	1-4									
Psychiatrist	0%											
Psychologist	8% (2/24)	2						6% (5)	2% ²			
Counsellor	8% (2/24)	1.5	1-2					4% (3)				
Social worker	37% (9/24)	1.8	1-3					7% (6)				
Care assistant/home car nurse	71% (17/24)	1.6	1-7					25% (20)	1% ²			
Wheelchair clinic	79% (19/24)							4% (3)				
Shoe clinic	4% (1/24)	2										
Hearing aid clinic	12% (3/24)	2										
Hydrotherapy	4% (1/24)	52										

Key: ¹ Living in Britain Survey 2001. ² British Household Panel Survey

It is notable that more children and young people in the ventilator-dependent sample used considerably more hospital services, primary care services and community health services than in the general population and a sample of severely disabled young people with hemiplegic cerebral palsy (Table 6.4). It was difficult to make specific comparisons between the service use of children under 15 years as little normative data exists.

NHS Services

In-patient care

Seventy-five percent (18/24) of ventilator-dependent children and young people living at home had been admitted for in-patient care during the past 12 months for a total of 367 days (mean number of admissions 1.8; range 1-4). This compares with 9% of children age 0-4 years, 4% of children age 5-15 years, 7% of young adults age 16-44 and 12% of young people with hemiplegic cerebral palsy. Ventilator-dependent children were mainly readmitted because of acute infections or a sudden deterioration in their condition; one child was readmitted for insertion of a phrenic nerve pacing device, which is a new technology for children who are dependent on ventilation 24 hours per day.

Out-patient care

All ventilator-dependent children attended either an Accident and Emergency department or out-patient appointment in the last 3 months compared with 13% of children age 0-4 years, 9% of children age 5-15 years, 12% young adults age 16-44 and 38% of young people with hemiplegic cerebral palsy.

Twenty-three ventilator-dependent children (96%) had a total of 127 out-patient appointments to see a doctor in the previous 12 months (mean number contacts 5.5; range 1-11).

(Note that data in Table 6.4 refers only to 3 months to facilitate comparisons with the population surveys.)

Primary care services

General Practitioner

A similar proportion of ventilator-dependent children (62%) had contact with their General Practitioner compared with young adults age 16-44 in the general population (72%) and those with hemiplegic cerebral palsy (74%). Ventilator-dependent children however had more contacts (mean 7.7) compared with young adults with hemiplegic cerebral palsy (mean 3), children age 0-4 years (mean 6) and children age 5-15 years (mean 3).

Dentist

All 24 ventilator-dependent children (100%) visited the dentist or orthodontist each year (mean 2 contacts per child), compared with 55% of young adults age 16-44, and 32% of young adults with hemiplegic cerebral palsy.

Health Visitor

Seven ventilator-dependent children who lived at home fell into the age group of children managed by a health visitor and all were visited during the previous 12 months (mean contacts 6.2, range 1-12). No population-based data could be found concerning the frequency of contact by Health Visitors with pre-school children.

Health services delivered in an educational setting

Eleven out of 13 (85%) ventilator-dependent children who attended school had been assessed by the school doctor in the past year. All 13 ventilator-dependent children (100%) who attended school had contact with the school nurse (mean contacts 82; range 1-190). Many of these children needed support with taking medication or enteral tube feeding whilst at school and required close supervision.

Community health services

All 24 ventilator-dependent children had a lot of contact with physiotherapists, speech therapists, occupational therapists, dieticians, psychologists etc. (total contacts: 1164, mean 48.5, range 1-190). For some ventilator-dependent

children therapy was received from health service staff that were based at their school. The most used services were physiotherapy (62%), speech or occupational therapy (50%), and the multidisciplinary wheelchair service (79%). Normative population-based data could not be found for children under sixteen years of age. It is difficult to make comparisons with the service use of young adults with hemiplegic cerebral palsy and young adults in the general population as the developmental needs of young children are different, with intensive input by therapists being concentrated at an earlier age to develop language and motor skills.

Pharmacy

Eighty-three percent of children (20/24) who lived at home required prescriptions for pharmaceutical products during the previous 12 months. Almost all of these children required regular repeat prescriptions for large amounts of pharmaceutical supplies, including: enteral feed preparations (9); oxygen (15); antibiotics (20); condition-specific medications and nebulizers (20). Many children used liquid versions of the medicine, which were more expensive and had a shorter shelf life. No population-based data could be found to make comparisons.

Equipment

All of the children living at home required a substantial amount of medical and health-related equipment. The main items that children used included: between 1 and 3 ventilators (24); gas humidifiers (22); oxygen monitors (21); resuscitation equipment (21); suction machines (19); manual/electric wheelchairs (20); power packs (19); hospital style electric or manual beds and mattresses (19); electric enteral food pumps (9); electric/manual hoists (9); oxygen concentrators (8); nebulizers (5); phrenic nerve pacers (3); and electricity generators (4). In addition, some children had such things as standing frames, blood pressure monitors, adapted buggies, chairs and seating (including car seats), braces, body casts and splints. Electrical items of equipment were usually covered by an annual service contract.

Disposable supplies

All children used large volumes of disposable equipment and supplies, although resource use varied depending on the complexity of the child's needs and underlying medical condition. Examples of disposable supplies included, ventilator circuits, tracheostomy tubes, face masks, bacterial filters, probes and sensors, various sizes of plastic connections and tubing, syringes, enteral feeding circuits, hypodermic needles, naso-gastric and gastrostomy tubes, incontinence supplies, urinary and suction catheters, sterile and disposable gloves, lotions, fluids and soaps, disposable hand towels, bedding, dressings and tapes etc.

Satisfaction with equipment and supplies

Twenty-four parents who had cared for their children at home during the last 12 months and 4 parents who had cared for their children for part of the year following discharge from hospital were asked if they had experienced any problems regarding equipment and disposable supplies. Over 60% (17/28) reported significant problems, with the main difficulty being interruptions to the supply and delivery of items to the home and the lack of product information.

Respite services

Fifteen families with ventilator-dependent children living at home accessed respite care services, including: social services family link scheme (2); social services residential facility (1); charity (4); NHS babysitting service (2); hospice (6). Nine families did not use respite services; of which, 3 had their own employed carers and did not have a need for respite; 2 did not want respite care and the remaining 4 had not been offered any.

Twenty families responded to a question regarding their feeling about the respite services available to them or their child. Five respondents (25%) were either completely or mostly satisfied; 5 (25%) were mixed, and 10 (50%) were either mostly or completely dissatisfied (mainly due to the lack of a suitable service and insufficient hours offered).

Service use of 7 ventilator-dependent children living in hospital in the previous 12 months

All health-related care and services used by ventilator-dependent children were included in the cost of a bed per 24 hours (apart from use of ambulances and visits to community health facilities). Children in hospital accessed the multi-disciplinary wheelchair clinic most frequently (range 1-3 visits) for the measuring and fitting of a wheelchair.

Ambulance services used by all 35 ventilator-dependent children in the previous 12 months

Fifty-seven percent (20/35) of ventilator-dependent children had used an ambulance in the previous 12 months. Use ranged from 1-10 trips per child (total 59 journeys). Almost all ambulances carried paramedics (52 journeys), 5 missions were carried out by the air ambulance and 2 journeys were carried out by a non-paramedic ambulance.

Education services used by 29 school aged ventilator-dependent children in the previous 12 months

Twenty-nine children were of school age or receiving further education. Of this group a variety of educational facilities were used: hospital school (3); combination of hospital and local education authority schools (3); mainstream primary (8); special primary (6); mainstream secondary (2); special secondary (3); home tuition (1); college of further education (2) and modern apprenticeship (1).

In the past 3 months of the school or college term, children and young people missed on average 18 days education (SD 18.74; range 0-60 days). This was mainly due to sickness of the child, sickness of the carer or classroom assistant, attendance at therapy or hospital out-patient appointments.

Additional educational support services

Nineteen children had a statement of educational need. Almost all of the children who had daytime employed carers provided by the NHS were cared for in school by a member of their own care team. Thirteen out of 29 children

required a taxi or minibus service to transport them, their equipment and their carer to school. Thirteen children had a full-time classroom assistant to help with their learning and participation in the classroom; 2 classroom assistants were also used by other children. Two children had part-time designated classroom assistants; 1 had a part-time note taker; and 1 had a break-time assistant. Three children had laptops and switches and 1 child had a communication device. A full-time nursery nurse was provided for 1 child and a part-time carer for 1 child. Additional health-related equipment was supplied for 3 children to use in school.

Satisfaction with education services

Just over half of respondents (59%; 17/29) were satisfied with their overall educational provision. Sixty-five percent (19/29) were satisfied or completely satisfied with their or their child's attendance at school. Three respondents (10%) had mixed feelings and 7/29 (25%) were either dissatisfied or completely dissatisfied. Those who were dissatisfied with education services tended to be respondents who had to overcome considerable barriers to try and access the right school and additional support for their child and had to cope with frequent absences of staff which negatively affected attendance.

Just over half of respondents (55%) said that they or their child currently had educational needs that required addressing. These needs ranged from wanting more education generally, to more specific input with Maths and English, additional classroom support to help the child integrate, and communication devices to enable the child to engage with the teacher and class.

Designated employed carers used by all 35 ventilator-dependent children in the previous 12 months.

Twenty families had designated employed carers to care for their children at home. This number included 2 children who were living in hospital and had their own care team in place prior to discharge. In addition, 2 children in hospital were waiting for carers to be recruited.

Different configurations of service use and type were noted, including: night-time care only (8); day-time care only (1); 24 hour care (11). Six children had teams of all qualified nurses or nursery nurses; 4 teams were made up of unqualified carers who had a qualified nurse supervising them from a distance; and 10 teams were made up of both qualified and unqualified nurses with varying ratios of qualified and unqualified staff. Most nurses and carers were supplied by the NHS, however some carers were provided via social services and the voluntary sector. Care teams were mostly supplied and managed by one sector (primarily the NHS), but some parents had accessed employed care from all 3 sectors of their own volition (NHS, social services and voluntary).

Satisfaction with employed care teams

Eighty-four percent (16/19) said that they had experienced problems with their care teams over the past 12 months. Problems cited were mainly due to the length of time taken to recruit carers, frequent sickness and absence of carers, personality clashes, unsuitable carers, lack of communication, rigid management policies and the constant threat of reduced hours.

A mixed picture emerged concerning respondent's feelings about the overall effectiveness of their or their child's care team in terms of meeting their needs. Forty-four percent (8/18) said that their care team was either completely or mostly effective; 39% (7/18) had mixed feelings; and 16% (3/18) said their care teams were either mostly or complete ineffective.

Social services used by all 35 ventilator-dependent children in the previous 12 months

Most of the support received by ventilator-dependent children from social services departments was from individual social workers who were heavily involved initially in helping to coordinate the discharge of the child from hospital. In addition, 2 children under 5 years of age who lived at home attended a social services nursery part-time and 1 had a nursery nurse. One family had a home help provided by social services. The relatively small amount of social services

provision concerning designated employed carers and respite care have been included under the respective sections.

Voluntary services and charitable resources used by all 35 ventilator-dependent children in the previous 12 months

Two families were helped to purchase an expensive wheelchair by a charity called Whizz-kidz. Other services used were mainly respite care (primarily hospice provision) and a small amount of designated employed care in the home. One charity also provided psychological services.

Independent funding

One family said that their current employed care team and specific items required by their child were paid for by an interim insurance payment.

Parental funding

One family had purchased an expensive piece of equipment for their child that was not readily available in the UK. A small number of parents had also contributed towards wheelchairs.

Satisfaction with the coordination and management of services used by all 35 Ventilator-dependent children in the previous 12 months

Fifty-seven percent (20/35) were either completely or mostly satisfied with the total amount of care and services received; 34% (12/35) had mixed feelings, and 9% (3/35) were either mostly or completely dissatisfied.

Forty-five percent (16/35) were either completely or mostly satisfied with the overall coordination of all of the care and services they received; 37% (13/35) had mixed feelings; and 16% (6/35) were either mostly or completely dissatisfied.

Those who were satisfied had a well managed care package (often with a team leader or case manager) and access to well-coordinated service provision that was regularly reassessed. Those who were dissatisfied tended to have the

opposite experience to those who were satisfied and in particular discharge management was highlighted as being ineffective and a major source of angst.

Review of care package

Of the 28 children who had lived at home for all or part of the previous 12 months, over 70% (20/28) had had their care package reviewed. For 8 families, no review had been undertaken; 4 of these families had not yet been home 12 months and the remaining 4 families could not remember the last time their care package had been reviewed.

Unmet needs

Health

In terms of health state, one ventilator-dependent child had been very unwell in the previous 12 months; 4 children (11%) said that they had been mainly unwell; 9 (26%) described their health as mixed and 22 (62%) said that they had been either mostly or completely well.

Just under a third of children (11/35) said they had health needs that required addressing, including the need for more named services or unresolved health problems. Children wanted more physiotherapy (6), occupational and speech therapy services (5), communication aids (3), 5 families wanted access to either more provision or more appropriate types of respite care and 5 families wanted more employed care. Two children were waiting for phrenic nerve pacers to be implanted.

Forty-eight percent of children (17/35) needed additional items of equipment, including 7 children who required new manual/powering wheelchairs.

Social

Thirty-five parents or, where appropriate, ventilator-dependent children were asked about the ventilator-dependent child's social life. Forty percent (14/35) were either completely or mostly satisfied, a further 40% (14/35) had mixed feelings, and 20% (7/35) were either mostly or completely dissatisfied. The

latter group mainly being those children in hospital who were socially isolated. Over 71% (25/35) reported issues regarding the child's social life that needed sorting out. These issues included wanting to meet and socialise with non-disabled children more often and the lack of activities and facilities for children with complex needs.

Transition to adult services

Thirty percent of young ventilator-dependent people over 13 years of age (3/10) had discussed the arrangements about transferring to adult services; 2 (20%) had already transferred; and for 5 young people (50%) no discussions had occurred.

The costs of support

The total costs of support will be presented under 4 sub headings:

- The entire group of 35 ventilator-dependent children;
- 7 children living in hospital for the previous 12 months;
- 24 children living at home over the last 12 months, and
- Comparison of hospital versus home care for 4 children discharged during the last 12 months.

The total costs of support for the entire group of 35 ventilator-dependent children

Of this group of 35 children, 24 lived at home, 7 lived in hospital and 4 were transferred home during the previous 12 months. Table 6.5 provides a summary view of the comprehensive costs of support received by all 35 children wherever their location (excluding the cost of adapting housing for a disabled child and the cost of domestic transportation).

The total cost of the support received by 35 ventilator-dependent children and young people was over £6.2 million with 92% of costs borne by the NHS, 3.2%

by education authorities, and 1% by social services (range £10,008 to £633,732 per child per year; SD £167,043).

Table 6.5. Summary view of the comprehensive costs of support received by all 35 children wherever their location.

<u>Annual support costs of all 35 children and young people:</u>		
NHS	£5,765,337	91.8%
Social services	£63,896	1.0%
Education	£200,902	3.2%
Charity/Voluntary services	£48,276	0.8%
Independent	£188,242	3.0%
Parents	£12,616	0.2%
Total	£6,279,269	100.0%

Cost category N=35	Sum	Range of costs (Min)	Range of costs (Max)	Mean cost	Standard Deviation	% total cost
7 hospital 4 hospital/home 24 home						
<u>Equipment *</u>						
NHS	£237,878	£1,894	£20,186	£6,797	£3,813	
Charity/Voluntary services	£5,606	£2,548	£3,058	£2,803	£361	
Parents	£12,616	£6,283	£6,333	£6,308	£36	
Independent	£7,116					
Total	£263,216	£1,894	£22,523	£7,520	£831	4.2%
<u>Hospital services</u>						
In-patient stays ~	£3,725,254	£0	£569,500	£106,436	£170,521	
Outpatient appointments	£17,346	£0	£1,665	£496	£406	
A&E attendances	£750	£0	£225	£21	£47	
Total	£3,743,350					59.6%
<u>Ambulances</u>	£19,649	£0	£5,000	£561	£1,007	0.3%
<u>Community health services #</u>						
NHS	£79,716	£72	£10,080	£2,278	£2,404	
Charity/Voluntary services	£1,206					
Total	£80,922					1.3%
<u>Primary care services +</u>	£23,344	£0	£3,776	£667	£734	0.4%
<u>Social services **</u>	£44,159	£0	£22,282	£1,206	£3,873	0.7%
<u>Pharmacy ***</u>	£54,822	£0	£10,556	£1,566	£2,447	0.9%
<u>Disposable equipment and supplies ****</u>	£223,653	£1,000	£15,876	£7,712	£5,287	3.6%
<u>Education++</u>	£200,902	£0	£18,050	£5,740	£6,286	3.2%
<u>Nursing care /personal care/respice ~</u>						
NHS	£1,382,925	£0	£158,369	£39,512	£54,669	
Social services	£19,737	£1,572	£13,871	£4,934	£5,966	
Charity/Voluntary services	£41,464	£242	£15,347	£5,183	£4,605	
Independent	£181,126					
Total	£1,625,252	£0	£181,126	£46,436	£58,461	25.9%
Grand Total	£6,279,269	£10,008	£633,732	£179,408	£167,043	100.0%

Key to Table 6.5.

* Equipment – All items of electronic and non- electric equipment (for example ventilators and standing frames).

~ In-patient stays – Costs used: Mean cost of intensive care and high dependency bed per day outside of London derived from NHS Finance Manual and 11 fieldwork sites; Mean cost of children's ward derived from Netten and Curtis (2002). In-patient costs per day cover all NHS services delivered in hospital apart from ambulance transport.

Community Health Services – For example chiropody, occupational therapy, physiotherapy, speech therapy, psychiatry, psychology and counselling.

+ Primary Care Services – General Practitioner, Opticians, Dentist.

** Social services – including social work input, social services nurseries etc.

*** Pharmacy – including all items available on prescription, including drugs, enteral feeds, oxygen, lotions, etc.

**** Disposable equipment and supplies –for example ventilator circuits, tracheostomy tubes, disposable hand towels, bedding, dressings and tapes etc.

++ Education – excludes the basic cost of a school place, includes all additional support services (for example classroom support) and transport to and from school.

~~ Nursing care, personal care and respite – including nurses, carers, home helps, and respite care delivered both inside and outside of the home.

The total costs of support for 7 children living in hospital for the previous 12 months

Table 6.6 provides a summary view of the comprehensive costs of support received by 7 children and young people living in hospital over the previous 12 months prior to interview (excluding the cost of adapting housing for a disabled child and the cost of domestic transportation). These children were either living in an intensive care unit or high dependency unit for children dependent on ventilators.

The total cost of the support received by these 7 ventilator-dependent children and young people was over £3.3 million with 98% of costs borne by the NHS. The cost of caring for these 7 children in hospital constitutes 53% of the total cost of caring for all 35 children. The average cost of services used by each child was £482 259 for the previous 12 months (range £335,489 to £633,732; SD £100,576). The cost of in-patient care consumed 92% of the total resource use. Two of the children had their own employed care team learning to look after them in hospital before being discharged home, which came to £106,481 (3.1%). This meant that the period of time prior to the child's discharge from hospital was the most expensive as both ward nurses and the child's own carers were being employed at the same time.

All of the children required significant input from a social worker in order to organise their discharge home and one child was 'looked after' by social services; the costs of social services received were £37,945 (1%).

The other costs were due to the educational needs of the children whilst in hospital; one child was attending a mainstream primary school and required a taxi to school each day and additional support in the classroom; the costs of extra educational services received were £26,809 (0.8%).

Table 6.6. The total costs of support for 7 children living in hospital for the previous 12 months.

Annual support costs of 7 children and young people in hospital		
NHS	£3,304,777	97.9%
Social services	£37,945	1.1%
Education	£26,809	0.8%
Charity/Voluntary services	£0	0.0%
Independent	£0	0.0%
Parents	£6,283	0.2%
Total	£3,375,814	100.0%

Cost category N=7 hospital	Sum	Range of costs (Min)	Range of costs (Max)	Mean cost	Standard Deviation	% total cost
<u>Equipment *</u>						
NHS	£64,370	£5,356	£16,241	£9,196	£4,110	
Charity/VS						
Parents	£6,283					
Independent						
Total	£70,653	£5,356	£22,523	£10,093	£6,106	2.1%
<u>Hospital services</u>						
In-patient stays ~	£3,124,000	£298,300	£569,500	£446,286	£85,325	
Outpatient appointments						
A&E attendances						
Total	£3,124,000	£298,300	£569,500	£446,286	£85,325	92.5%
<u>Ambulances</u>	£5,211	£0	£2,565	£744	£906	0.2%
<u>Community health services #</u>						
NHS	£4,530					
Charity/Voluntary services						
Total	£4,530	£344	£1,032	£647	£226	0.1%
<u>Primary care services +</u>	£185					0.0%
<u>Social services **</u>	£37,945	£1,950	£22,282	£5,421	£7,554	1.1%
<u>Pharmacy ***</u>	included					
<u>Disposable equipment and supplies ****</u>	included					
<u>Education ++</u>	£26,809	£0	£13,739	£3,830	£5,791	0.8%
<u>Nursing care /personal care/respite ~~~</u>						
NHS	£106,481	£0	£48,803	£15,212	£19,418	
Social services						
Charity/VS						
Independent						
Total	£106,481	£0	£48,803	£15,212	£19,418	3.2%
Grand Total	£3,375,814	£335,489	£633,732	£482,259	£100,576	100.0%

Key to Table 6.6.

* Equipment – All items of electronic and non- electric equipment (for example ventilators and standing frames).

~ In-patient stays – Costs used: Mean cost of intensive care and high dependency bed per day outside of London derived from NHS Finance Manual and 11 fieldwork sites; Mean cost of children's ward derived

from NHS finance manual. In-patient costs per day cover all NHS services delivered in hospital apart from ambulance transport.

Community Health Services – For example chiropody, occupational therapy, physiotherapy, speech therapy, psychiatry, psychology and counselling.

+ Primary Care Services – General Practitioner, Opticians, Dentist.

** Social services – including social work input, social services nurseries etc.

*** Pharmacy – including all items available on prescription, including drugs, enteral feeds, oxygen, lotions, etc.

**** Disposable equipment and supplies –for example ventilator circuits, tracheostomy tubes, disposable hand towels, bedding, dressings and tapes etc.

++ Education – excludes the basic cost of a school place, includes all additional support services (for example classroom support) and transport to and from school.

~~ Nursing care, personal care and respite – including nurses, carers, home helps, and respite care.

The total costs of support for 24 children living at home for the previous 12 months

The total cost of the support received by these 24 ventilator-dependent children and young people was over £2.5 million (excluding the cost of adapting housing for a disabled child and the cost of domestic transportation) with 83% of costs borne by the NHS (Table 6.7). The average costs of services used by ventilator-dependent children and young people were £104,352 per child per year (range £10,008 to £331,619; SD £84,082). Children with the most expensive support packages had employed care teams with a high ratio of trained nurses and/or required readmission to hospital during the previous 12 months.

Providing employed care in the home or respite facility for ventilator-dependent children absorbed 55% of total costs (£1.33 million). Home care services were predominantly provided by the NHS (83%). Charities and hospices mainly provided respite care (13.5%), social services provided a mixture of respite and personal care in the home (1.4%) and independent finance (insurance awards) was used to purchase (13.5%) of nursing and personal care services.

The considerable health-related service use by this group of ventilator-dependent children was outlined in more detail in a previous paragraph and illustrated in Table 6.4. Hospital services absorbed 17.8% of the budget (£444,994) with 97% being spent on in-patient care. Hospital admissions were

mainly due to acute illness, but some ventilator-dependent children were admitted to hospital for major surgery and the implantation of new expensive technology, which aimed to improve their quality of life, but would not reduce their overall resource use.

The long-run marginal costs of purchasing and servicing equipment consumed around 7% of the total budget (£173,288). Equipment costs ranged from £1,894 to £20,186 per child per year (Mean £7,220; SD £4,689). Ventilator-dependent children consumed varying amounts of disposable equipment and supplies (range £1,000 – £15,876) with an average cost of £8,133 per child per year, which absorbed 8% of the total budget. Eighteen ventilator-dependent children and young people required additional support in the classroom and or transport to school; this absorbed 7% of the total budget (Mean £7,143 per child per year).

Key to Table 6.7

* Equipment – All items of electronic and non- electric equipment (for example ventilators and standing frames).

~ In-patient stays – Costs used: Mean cost of intensive care and high dependency bed per day outside of London derived from NHS Finance Manual and 11 fieldwork sites; Mean cost of children's ward derived from NHS Finance Manual. In-patient costs per day cover all NHS services delivered in hospital apart from ambulance transport.

Community Health Services – For example chiropody, occupational therapy, physiotherapy, speech therapy, psychiatry, psychology and counselling.

+ Primary Care Services – General Practitioner, Opticians, Dentist.

** Social services – including social work input, social services nurseries etc.

*** Pharmacy – including all items available on prescription, including drugs, enteral feeds, oxygen, lotions, etc.

**** Disposable equipment and supplies –for example ventilator circuits, tracheostomy tubes, disposable hand towels, bedding, dressings and tapes etc.

++ Education – excludes the basic cost of a school place, includes all additional support services (for example classroom support) and transport to and from school.

~~ Nursing care, personal care and respite – including nurses, carers, home helps, and respite care delivered both inside and outside of the home.

Table 6.7. The total costs of support for 24 children living at home over the last 12 months.

Annual support costs of 24 children and young people at home		
NHS	£2,076,343	82.9%
Social services	£21,088	0.8%
Education	£171,424	6.8%
Charity/Voluntary sector	£41,022	1.6%
Independent	£188,242	7.5%
Parents	£6,333	0.3%
Total	£2,504,452	100.0%

Cost category N=24 at home	Sum	Range of costs (Min)	Range of costs (Max)	Mean cost	Standard Deviation	% total cost
<u>Equipment *</u>						
NHS	£154,233	£1,894	£20,186	£6,426	£3,723	
Charity/Voluntary sector	£5,606	£0	£3,058	£234	£795	
Parents	£6,333					
Independent	£7,116					
Total	£173,288	£1,894	£20,186	£7,220	£4,689	6.9%
<u>Hospital services</u>						
In-patient stays ~	£429,918	£0	£154,700	£17,913	£35,724	
Outpatient appointments	£14,476	£0	£1,260	£603	£307	
A&E attendances	£600	£0	£225	£25	£53	
Total	£444,994			£18,541		17.8%
<u>Ambulances</u>	£11,019	£0	£5,000	£459	£1,030	0.4%
<u>Community health services #</u>						
NHS	£60,470	£72	£7,353	£2,520	£2,219	
Charity/Voluntary sector						
Total	£60,470					2.4%
<u>Primary care services +</u>	£20,788	£0	£3,776	£866	£789	0.8%
<u>Social services **</u>	£1,351	£0	£998	£56	£200	0.1%
<u>Pharmacy ***</u>	£48,179	£0	£10,556	£2,007	£2,741	1.9%
<u>Disposable equipment and supplies ****</u>	£195,183	£1,000	£15,876	£8,133	£5,216	7.8%
<u>Education ++</u>	£171,424	£0	£18,050	£7,143	£6,465	6.8%
<u>Nursing care /personal care/respite ---</u>						
NHS	£1,141,477	£0	£158,369	£47,562	£59,995	
Social services	£19,737	£0	£13,871	£822	£2,859	
Charity/Voluntary sector	£35,416	£0	£15,347	£1,476	£3,472	
Independent	£181,126					
Total	£1,377,756	£0	£181,126	£57,407	£63,710	55.0%
Grand Total	£2,504,452	£10,008	£331,619	£104,352	£84,082	100.0%

Ventilator-dependency related costs of support.

It was decided to estimate the extra costs of services for children dependent on ventilators, that is the costs associated with services used as a result of their impairment or combinations of conditions. All service use of the 24 ventilator-dependent children living at home was checked to see if any services could be attributed to a category other than their disability or underlying diagnosis.

All services were found to be attributable to their disabilities, although some basic healthcare and child health surveillance were combined with activities directly associated with the child's impairments and medical diagnoses. For example, ventilator-dependent children would have arguably been likely to have visited a dentist on an annual basis, but when probed children mainly required the services of a hospital-based dentist due to their requirement for specialist monitoring. Many of the children required additional dental work due to the ingestion of sugar-based liquid medicines up to 3 times a day and some children needed specialist braces and splints because ventilator masks caused some facial anomalies.

Ventilator-dependent children who were managed by health visitors either accessed a specialist health visitor dealing with disabled children, or for issues around continence. Only 62% of ventilator-dependent children saw their General Practitioner in the previous 12 months and the reasons given were always related to the child's impairments and medical conditions. Ventilator-dependent children who did not see their General Practitioner usually deliberately bypassed them and went directly to hospital, where their complex needs could be more adequately met; most families were well known to hospital services and had negotiated arrangements for direct access to a specialist doctor.

The extra costs associated with ventilator-dependency can be attributed to the total costs of support for the previous 12 months (mean cost of £104,352 per child per year). In contrast, Beecham *et al.* (2001) calculated the extra costs associated with hemiplegic cerebral palsy to be £5,600 per young adult per year at 1998-1999 prices. After adjusting prices to reflect inflation, the average cost of supporting a ventilator-dependent child at home was over 18 times greater than the average cost of managing a young person with hemiplegic cerebral palsy. The major differences in costs can be attributed to ventilator-dependent children's greater use of employed carers, health services and need for additional support services at school.

Costs of 'simple' ventilator-dependency versus 'complex' ventilator-dependency

Although all 24 children at home met the definition for ventilator-dependency, there was considerable variation in the extent to which associated diagnoses and disabilities affected them. Five children and young people were assessed as having less complex needs as they used mask ventilation at night, their life was not totally dependent on the technology, they did not have apparent learning difficulties, and they had minimal to medium difficulties with their mobility. For this group of children with 'simple' ventilator-dependency and moderate associated impairments, the average total support costs per child per year were £17,876. In contrast for the remaining 19 children and young people with more complex needs, the average total support costs per child per year was £127,109, over 7 times greater.

Comparison of hospital versus home care for 4 children discharged during the last 12 months

These 4 children provided the opportunity to calculate the costs of care received whilst in hospital and then compare the costs of their home care package (Table 6.8).

Their total annual support costs were £830,000. For all 4 children who were discharged home, an equivalent cost of support was calculated for a month of hospital care and a month of home care (excluding the cost of adapting housing for a disabled child and the cost of domestic transportation). Taking into account that for one child their employed care team spent 4 months in hospital learning to look after them, the actual cost of home care was up to 44% cheaper than hospital care. Two children were discharged with employed care teams (one 24 hour the other 3 nights per week, and both with a high ratio of unqualified carers). Two children were provided with regular respite care. One child was discharged home without any employed or respite care, to be cared for by his parents.

Key to Table 6.8

* Equipment – All items of electronic and non- electric equipment (for example ventilators and standing frames).

~ In-patient stays – Costs used: Mean cost of intensive care and high dependency bed per day outside of London derived from NHS Finance Manual and 11 fieldwork sites; Mean cost of children's ward derived from Netten and Curtis (2002). In-patient costs per day cover all NHS services delivered in hospital apart from ambulance transport.

Community Health Services – For example chiropody, occupational therapy, physiotherapy, speech therapy, psychiatry, psychology and counselling.

+ Primary Care Services – General Practitioner, Opticians, Dentist.

** Social services – including social work input, social services nurseries etc.

*** Pharmacy – including all items available on prescription, including drugs, enteral feeds, oxygen, lotions, etc.

**** Disposable equipment and supplies –for example ventilator circuits, tracheostomy tubes, disposable hand towels, bedding, dressings and tapes etc.

++ Education – excludes the basic cost of a school place, includes all additional support services (for example classroom support) and transport to and from school.

~~ Nursing care, personal care and respite – including nurses, carers, home helps, and respite care delivered both inside and outside of the home.

Table 6.8. Comparison of hospital versus home care for 4 children discharged during the last 12 months.

Comparison of support costs of 4 children and young people in hospital and at home								
	Child A Hospital 3 Months	Child A Home 9 Months	Child B Hospital 4 Months	Child B Home 8 Months	Child C Hospital 7 Months	Child C Home 5 Months	Child D Hospital 1.5 Months	Child D Home 10.5 Months
NHS	£73,942	£18,016	£139,265	£118,156	£363,558	£21,293	£73,992	£8,200
Social services	£25	£5,806	£0	£3,838	£975	£0	£25	£0
Education	£0	£2,669	£0	£0	£0	£0	£0	£0
Charity/Voluntary S	£0	£0	£0	£0	£0	£241	£0	£0
Independent	£0	£0	£0	£0	£0	£0	£0	£0
Parents	£0	£0	£0	£0	£0	£0	£0	£0
Total	£73,967	£26,491	£139,265	£121,994	£364,533	£21,534	£74,017	£8,200
Yearly Total	£100,458		£261,259		£386,067		£82,217	

Cost category N=4 Discharged home during previous 12 m	Child A Schoolchild Mask Ventilation at Night		Child B Toddler 24hr Ventilation via Tracheostomy		Child C Toddler 24hr Ventilation via Tracheostomy		Child D Schoolchild Mask Ventilation at Night	
	Hospital 3 Months	Home 9 Months	Hospital 4 Months	Home 8 Months	Hospital 7 Months	Home 5 Months	Hospital 1.5 Months	Home 10.5 Months
<u>Equipment *</u>								
NHS	£883	£2,649	£2,226	£4,452	£3,928	£2,804	£291	£2,235
Charity/Voluntary S								
Parents								
Independent								
Total	£883	£2,649	£2,226	£4,452	£3,928	£2,804	£291	£2,235
<u>Hospital services</u>								
In-patient stays ~	£72,796	£478	£96,000	£15,968	£357,000		£73,100	
Outpatient appointments				£505		£222		£1,665
A&E attendances				£75			£75	
Total	£72,796	£478	£96,000	£16,548	£357,000	£222	£73,175	£1,665
<u>Ambulances</u>	£263	£0	£0	£0	£2,630	£0	£526	£0
<u>Community health services #</u>								
NHS		£10,080		£1,750		£986		£1,899
Charity/Voluntary S								
Total	£0	£10,080	£0	£1,750	£0	£986	£0	£1,899
<u>Primary care services</u>		£570		£343		£734		£724
<u>Social services **</u>	£25			£3,838	£975		£25	
<u>Pharmacy ***</u>		£239		£4,265		£450		£677
<u>Disposable equipment and supplies ****</u>		£4,000		£8,720		£4,250		£1,000
<u>Education ++</u>		£2,669						
<u>Nursing care /personal care/respite ---</u>								
NHS			£41,039	£82,078		£11,847		
Social services		£5,806						
Charity/Voluntary S						£241		
Independent								
Total	£0	£5,806	£41,039	£82,078	£0	£12,088	£0	£0
Grand Total	£73,967	£26,491	£139,265	£121,994	£364,533	£21,534	£74,017	£8,200
Cost per Month	£24,656	£2,943	£34,816	£15,249	£52,076	£4,307	£49,345	£781
Yearly Total	£100,458		£261,259		£386,067		£82,217	

Comparison of total costs of support in different hospital and home care settings

The total costs of support in 12 care settings (3 hospital, 7 home with various types and levels of employed care, and 2 residential) were calculated using where appropriate mean costs from the present economic evaluation (Table 6.9). Costs for residential care were derived from Netten and Curtis (2002) and the National Hospital for Neuro-disability 'life-long home' ventilator unit at Putney (2002 prices).

The cost of living in hospital ranged from £630,388 (intensive care) to £155,158 (children's ward) per child per year. Costs of living at home ranged from £248,207 (24 hr care E grade nurses) to £46,483 (care by parent(s)) per child per year. The cost of a residential placement ranged from £105,555 to £194,351* per placement per year. * Placement in outer London and includes relevant supplements.

It was not always cheaper for a ventilator-dependent child to live at home; a care package including qualified nurses and 24 hour care (£248,207) was more expensive than a children's ward (£154,841) and residential placement (£105,555 to £194,351) but less expensive than a long-term ventilation unit (£301,571) and intensive care unit (£630,071) where most ventilator-dependent children live whilst in hospital.

Some ventilator-dependent children with less complex needs are already managed on children's wards where the ratio of staff to children is lower than in intensive care and high dependency units. An additional carer may be required on a children's ward to care for a ventilator-dependent child with complex needs. This will increase costs but is very unlikely to be more than £93,366, which would place care on a children's ward at the same level as 24 hour care at home by qualified nurses.

It was cheaper to employ unqualified carers at home who were managed by a qualified team leader. Shift enhancements meant that it was between x1.3 and x2 more expensive to employ staff at night, weekends and bank holidays.

Even if the lowest costs for intensive care (£1,300) are substituted for the average cost (£1,700) per 24 hours, the total cost of £474,500 is still more expensive than the total costs of support of all other packages. A minority of long-term ventilation units charged as little as £500 per day (£182,500 per child per year); these units were staffed by a high ratio of unqualified to qualified staff and would be cheaper than 24 hour home care packages with more than 50% qualified staff.

Key to Table 6.9.

* Equipment – Mean costs of equipment derived from 24 ventilator-dependent children living at home (Table 6.7). For children living in hospital, it was assumed that they used hospital equipment.

** Hospital services – Mean costs of all in-patient, out-patient and A&E services derived from 24 ventilator-dependent children living at home (Table 6.7). For children living in hospital, mean costs per day included all pharmacy, and disposable equipment and supplies.

Community Health Services – Mean costs of services derived from 24 ventilator-dependent children living at home (Table 6.7) and 7 children living in hospital (Table 6.6).

+ Primary Care Services – Mean costs of services derived from 24 ventilator-dependent children living at home (Table 6.7).

Social services – Mean costs of social services derived from 24 ventilator-dependent children living at home (Table 6.7) and 7 children living in hospital (Table 6.6).

*** Pharmacy – Mean costs of pharmacy derived from 24 ventilator-dependent children living at home (Table 6.7). For children in hospital, it was assumed that pharmacy costs were included in the cost of a bed per day.

**** Disposable equipment and supplies – Mean costs derived from 24 ventilator-dependent children living at home (Table 6.7). For children in hospital, it was assumed that pharmacy disposable equipment and supplies were included in the cost of a bed per day.

++ Education – Mean costs of additional education services derived from 24 ventilator-dependent children living at home (Table 6.7) and 7 children living in hospital (Table 6.6).

-- Nursing care and personal care – For children living in hospital, nursing care costs were included in the cost of a bed per day. Home care, costs were derived from Netten and Curtis, (2002): Shift allowances were paid as follows: For NHS staff: weeknights and Saturdays x1.3 and Sundays/bank holidays x1.6. For local authority staff, Saturdays x1.5, week nights x1.3, Sundays/bank holidays x2. Weekly costs of residential care were provided by the National Hospital for Neuro-disability, Putney whose costs include London W8ing and London Supplement, and Netten and Curtis, (2002).

Table 6.9. Comparison of total costs of support in different hospital and home care settings.

Model	A	B	C	D	E	F	G	H	I	J	K	L
Package	12 months Paediatric intensive care unit @£1700 per 24 hrs	12 months High dependency Long term ventilation unit@£800per 24 hrs	12 months Children's ward @£398 per 24 hrs	12 months 24 hour home care E grade nurses £184 987 Team leader Grade F Nurse 0.2 WTE £8 385	12 months 24 hour home care 50% E grade nurses £92,494 B grade HCA £53 153 Team leader Grade F Nurse 0.2 WTE £8,385	12 months 24 hour home care B grade HCA £106,306 Team leader Grade F Nurse 0.2 WTE £8,385	12 months 24 hour home care LA home care worker £101,708 Team leader Grade F Nurse 0.2 WTE £8,385	12 months 12 hour night home care B grade HCA £57,583 Team leader Grade F Nurse 0.2 WTE £8,385	12 months 12 hour day care B grade HCA £48,723 Team leader Grade F Nurse 0.2 WTE £8,385	12 months Parent unpaid carers	12 months High dependency nursing home for disabled young adults@ £1,136 per week	12 months 'Home for Life' Royal Hospital for Neuro-disability' Ventilator Unit for young people and adults @ £3000 per week for basic care in outer London
Nursing and personal care				£193,372	£154,032	£114,691	£110,093	£65,967	£57,108	£0	£59,072	£156,000
Equip ment *				£7,220	£7,220	£7,220	£7,220	£7,220	£7,220	£7,220	£7,220	£7,220
Hospital services **	£620,500	£292,000	£145,270	£18,541	£18,541	£18,541	£18,541	£18,541	£18,541	£18,541	£18,541	£18,541
Community health services #	£647	£647	£647	£2,519	£2,519	£2,519	£2,519	£2,519	£2,519	£2,519	£2,519	£2,519
Primary care services +				£866	£866	£866	£866	£866	£866	£866	£866	£866
Pharmacy ***				£2,007	£2,007	£2,007	£2,007	£2,007	£2,007	£2,007	£2,007	£2,007
Disposable equipment and supplies****				£8,132	£8,132	£8,132	£8,132	£8,132	£8,132	£8,132	£8,132	included
Social services ##	£5,412	£5,412	£5,412	£56	£56	£56	£56	£56	£56	£56	£56	£56
Education ++	£3,829	£3,829	£3,829	£7,142	£7,142	£7,142	£7,142	£7,142	£7,142	£7,142	£7,142	£7,142
TOTAL	£630,388	£301,888	£155,158	£239,855	£200,515	£161,174	£156,576	£112,450	£103,591	£46,483	£105,555	£194,351

6.4 Discussion

Demographic issues

In the sample two thirds of ventilator-dependent children were males; this is due to the group of genetic disorders such as muscular dystrophy, which only occur in males. This over-representation of males is also reflected in other studies of childhood disability (Perrin, 2002).

Seventy-five percent of families had a yearly income that was less than the average wage in 2002, and above average levels of adult unemployment were found. This finding is replicated in numerous other studies on welfare and income amongst families with disabled children.

Maternal and paternal levels of educational qualifications were at both extremes of the spectrum, with more mothers and fathers having achieved no qualifications than in the general population and higher levels of both parents being awarded a diploma or a degree.

Long-run marginal services costs of supporting ventilator-dependent children and young people

The costs of long-term ventilation arose from decisions to commit (or not commit) resources to the treatment of underlying disease and to interventions to minimise the disabling barriers that resulted from impairments. The focus of this economic evaluation has been on the unit costs of providing formal health, social, educational and voluntary services. Data presented in this chapter have begun to identify the long-run marginal service costs of supporting ventilator-dependent children and young people over 12 months in hospital and at home. Findings revealed that the entire group of 35 ventilator-dependent children and young adults consumed services and resources costing over £6.2 million during the preceding 12 months.

These costs, however, are likely to be underestimated, especially as housing adaptations and domestic transportation costs were not included in the

calculations. The exclusion of housing costs, in particular, will have an inevitable impact on the total cost of social service provision. Many houses had installed railings, adapted bathrooms and ceiling hoists that were fitted with the aid of a means tested disabled facilities grant. Individual client data and additional data derived from Beresford and Oldham, (2002) indicate that, as the disabled facilities grant is means tested and capped at £20,000, even if all 35 children received the maximum grant and the adaptations only lasted 5 years, the cost per annum (£140,000) would amount to 2.2% of total annual support costs of just over £6.2 million.

Excess costs of ventilator-dependency

Ventilator-dependent children arguably have the most complex needs of all children with disabilities. Using the CSRI, it was possible to compare the support costs of ventilator-dependent children with normative data and other groups of children in order to give an order of magnitude estimate of the 'excess' costs. Findings revealed that ventilator-dependent children consumed up to 18 times more resources than a population of young adults with hemiplegic cerebral palsy and where comparable data were available, they consumed considerably more resources than children in the general population.

New technology

Two children included in the economic evaluation had a new technology called a phrenic nerve pacing device implanted; one child had the device inserted during the previous 12 months. This new and expensive technology added to the costs of care and resources for the children concerned as the phrenic nerve pacer was used in addition to their existing ventilators. For the child who had the device implanted during the previous 12 months the costs associated with the device were as follows: 3 month in-patient stay in a spinal injuries unit (£35,820); long-run marginal cost of phrenic nerve pacing device (£6,670) per year; and, disposable supplies per year (£500).

The cost of hospital versus home care

Like-for-like comparisons between hospital and home care revealed that for most children and young people care at home was cheaper than in hospital, but this depended on the complexity of the child's needs, the amount of qualified nursing care required at home, the number and duration of readmissions to hospital and the ability of parents to provide unpaid care. Findings revealed that for children with 'simple' ventilator-dependency who did not need employed carers at home; living at home was always cheaper than living in hospital. For children with 'complex' ventilator-dependency and who needed 24 hour care at home by qualified nurses, it was not always cheaper living at home with a package of support services compared with living in a children's ward or residential home. This mirrors evidence from the United States where legislation was introduced to eliminate 24 hour home qualified nursing care as it was more costly to Medicaid than long-term ventilation units, where many people on ventilators could be cared for by a small team of staff (Bach *et al.* 1992). Unlike in the United States, unqualified carers in the UK are not prevented by law from undertaking 'nursing' interventions and as such unqualified carers have increasingly been employed to provide nursing and personal care in the home.

The Medicaid policy preference of institutional care did not, however, take into account the potential benefits or disbenefits of institutional versus home care or provide patients with a choice of where they wanted to live. The impact of the child's environment on their health-related quality of life was explored previously in Chapters 3-5. In Chapter 9, evidence will be constructed concerning the costs of caring for children in different settings compared with the social benefits (consequences) to determine which placements confer the desired psychosocial outcomes from the perspective of the children and their families.

Costed packages of care

Case study data was used with good effect to build up examples of costed packages of care, which may be used as a resource by commissioners, managers and parents. Findings were circulated to doctors, commissioners

and managers for verification. One doctor said that he had deliberately declined funding to build a long-term ventilation unit at his hospital as he considered that the lower costs associated with such a unit would act as a disincentive to funders to purchase a package of home care; figures derived from costed packages of care provided evidence to support his view.

Satisfaction with services

Taken as a whole, around half of children and young people had well coordinated and effective packages of support, and the other half had elements or entire packages that were disorganised and ineffective. Overall, 57% of parents and children were satisfied with their total package of care, and slightly fewer (45%) were satisfied with the coordination and management of their total care package. There were specific areas of dissatisfaction with aspects of care packages (discharge management, logistical issues around delivery of supplies, problems with employed care teams, fragmented service provision, poor management and coordination, and lack of appropriate respite care). Around a third of children had health needs that required resolving, and a similar proportion wanted additional items of equipment. Sixty percent of children indicated that their care packages failed to provide interesting/fulfilling social lives and around half of children in full-time education said that they needed additional services in order to engage in learning more fully.

Parents who were the most dissatisfied were those whose children in the main had the most unmet needs and who had either lived in hospital during the previous 12 months or were parents of children with 'complex' ventilator-dependency who lived at home and had multifaceted packages of care which were managed poorly. Interestingly, the parent with the most expensive care package of all the children living at home, who also lived in a state of the art purpose-built local authority house, was dissatisfied because of the area that they lived in. The issue of dissatisfaction around high cost complex care packages will be discussed further in Chapter 7.

Parents' opinions around satisfaction may be influenced by unrealistic expectations of either the ventilator-dependent person, their family or the services (Beecham *et al.* 2001). The quality of services and service delivery may also have an impact on the subjective views of unmet need. Despite limitations in interpreting data on satisfaction and unmet need, it does provide an indication of the adequacy of support and therefore of the associated costs; which for around half of children and young people would be greater if they received their desired care package.

Strengths and weaknesses of the CSRI

One of the main strengths of this study was the methodological rigour with which it was carried out, which enabled new rigorous evidence around costs to be produced. During interviews, a detailed survey of service and resource use was obtained which was supplemented by complete inventories of resource use provided by some parents. If parents or young people were not able to provide information on health, social or educational services, consent was obtained to approach a relevant professional. Unit costs were attributed primarily from a national compilation of unit costs that were calculated using a validated methodology.

Data input and calculations were verified by a second checker to ensure accuracy. The results were fed back at an early stage to members of the UK Paediatric Long-Term Ventilation Working Party. Feedback confirmed that the range of in-patient costs matched with their clinical experience. Individuals expressed surprise at the cost of some home care packages, but this was explained by the fact that unit costs per hour of service were more expensive than the hourly rate of a professional, and the total cost of care included health, social, educational and voluntary services. Although the published studies that estimated costs of hypothetical ventilator-dependent children in the UK had methodological limitations, taking inflation into account, the costs of employed carers and disposable equipment and supplies fell within the range of equivalent costs in this study.

This method of economic evaluation has a number of criticisms that warrant discussion. Primarily, the costs represent services currently provided or used by children and young people. These data do not discriminate between over or under provision that produce excessively high or low costs, or provide explanations as to whether expenditure generated good outcomes for ventilator-dependent children or their families (Beecham *et al.* 2001).

Nor does this method estimate the cost of hidden work undertaken by professionals. It is highly likely that other than face-to-face contact, professionals spend time on issues concerning the child and their family. One such example would be the time spent and costs associated with recruiting a team of carers.

The range and average costs of in-patient care were obtained from the NHS Finance Manual and from participating fieldwork sites. These costs were calculated by dividing the total annual budget of the ward or intensive care unit by the number of beds and 365 days to come up with a daily cost of a bed. This method does not determine actual resources used by individual patients. Edenbrook *et al.* (1997) have piloted a new activity-based costing method for accurately identifying individual patient costs in intensive care, but as yet this method has not been used widely in economic studies. Edenbrook *et al.* calculated an average daily cost of intensive care per patient in 1995 of £1,148 with a standard deviation of £243. The narrow range of costs was accounted for by the fact that all patients in intensive care whatever their diagnosis needed the same basic things. Costs were only significantly higher for the first 24 hours of care, thereafter there was no significant difference between costs of care for subsequent 24 hour periods. Allowing for inflationary pressures over 8 years, the mean cost of £1,700 per day used in this study appears to be within a comparable range.

Additional data around satisfaction and unmet needs aimed to assess adequacy and appropriateness of current services and this information helped to place service provision within an individual context. There are however, no evaluations of interventions, treatments or models of service delivery for

children and young people who are dependent on ventilators. Thus no evidence exists on which to make a judgment as to which services might constitute an optimal care package with the desired outcomes for the child and their family.

In full economic evaluations, Knapp (1993), and Byford and Rafferty (1998) recommend taking a societal perspective including the costs to society at large as well as those benefiting from the intervention. As mentioned previously, it was originally the intention to undertake this study from a societal perspective, but like the experience of Netten and Curtis (2002), this information was found to be difficult to collect and generated an overly time consuming task of attributing costs, especially concerning the input of parents and relatives. Few published economic evaluations include the input of informal carers and as such it was not possible to make direct comparisons with studies using a similar methodology. Although beyond the limitations of this study, the contribution of parents and relatives is important, as their ability to care for their child on their own resulted in significant savings to the taxpayer. Carers UK calculated the value of carers' support in 2002 at an hourly rate of £9.95 per hour (Carers UK, 2002).

Synthesised findings derived from the 4 research themes will be reviewed in an evidence based and policy context in Chapter 9 and further research directions will be outlined in Chapter 10.

6.5 Summary

Chapter 6 focused on the costs of caring for children dependent on ventilators (theme 3), and satisfaction with services received (theme 4).

Theoretical and legal imperatives around the allocation and distribution of finite resources were explored; the successful use by parents of the Human Rights Act as a lever to access previously denied resources was developed further from previous chapters.

Findings from a study evaluating the costs of care and services received in the previous 12 months by 35 children and young people dependent on ventilators were reported. The economic study aimed to estimate the long-run marginal service costs of supporting ventilator-dependent children in hospital, home and residential settings over 1 year; levels of satisfaction with services received were also measured and gaps in service provision estimated. Total annual support costs to the state and voluntary sector of supporting 35 ventilator-dependent children were estimated to be over £6.2 million. It was mainly but not always cheaper for the child to live at home. Around half of children were satisfied with the care and services received; substantial gaps were identified in service provision especially around social outcomes.

These data provide new evidence concerning the costs of caring for ventilator-dependent children. The chapter concluded by comparing findings with published literature and discussing the strengths and weaknesses of the study.

The next chapter continues to focus on the costs of care (theme 3) and satisfaction with services (theme 4) with qualitative methods.

Chapter 7

Exploring and explaining individual patient and family costs and satisfaction with services using qualitative methods: Is the burden of care on families reasonable?

7.0 Introduction

In Chapter 6, the total costs of formal services received and resources used by children dependent on ventilators was reported. These costs however, did not include the costs to families of caring for a child dependent on a ventilator. In order to redress this imbalance in perspective, this chapter continues to focus on the costs of caring (theme 3) and satisfaction with services (theme 4) by exploring the costs of care, in particular those incurred by families, and satisfaction with services through qualitative methods.

The chapter begins by reviewing the few qualitative studies around costs and satisfaction with services that have been undertaken with children dependent on ventilators. A qualitative study underpinned by the social model of disability that aimed to explore and explain individual and family costs and satisfaction with services is then reported.

7.1 Exploring and explaining costs and satisfaction with services using qualitative methods

In Chapter 4, the relative merits of undertaking qualitative research to complement quantitative findings were outlined. It was decided to undertake a qualitative study to complement the quantitative economic data described in Chapter 6, and then undertake methodological work to synthesise both quantitative and qualitative cost data around exploring and explaining costs of

caring for children dependent on ventilators, which will be described in Chapter 8.

What is already known about the costs of caring for children dependent on ventilators, and satisfaction with services: A summary of the qualitative literature.

At the beginning of the empirical work reported here, Sudbury and Noyes (1999) published a comprehensive review of the literature which summarised the considerable negative economic consequences for the family and primary caregivers of caring for a disabled child generally, and when literature was available, concerning the ventilator-dependent child specifically. A summary of key issues is presented here, for a more in-depth discussion reference can be made to Sudbury and Noyes (1999).

General financial pressures on families

Severe financial pressures around caring for a disabled child generally were documented as arising from a whole range of sources (Baldwin and Carlisle, 1994; Beresford *et al.* 1996), including: the inability of at least one parent to take paid employment; the costs of special travel arrangements; the need to purchase specialised varieties of a whole range of items of which others purchase a standard version; for some, clothing and laundry costs; and individual circumstances which necessitated a host of different special arrangements. In the review by Glendinning and Kirk (1998), parents expressed angry frustration at the substantial state expenditure on unsuitable equipment for their child (often in bulk purchase) whilst they were left to pay privately for equipment, which would be more suitable.

Parents of technology-dependent children who took part in focus groups (Diehl *et al.* 1991) identified finance as one of the significant categories of need. Financial issues around providing suitable housing were amongst the most debilitating and widespread pressures reported in the more general UK reviews of literature around childhood disability (Baldwin and Carlisle, 1994; Kirk, 1998; Kirk and Glendinning, 1998). The United States literature varies in the

identification of housing as an issue; the editorial by Orlowski (1993), which gave a concise overview of the whole subject, took for granted that readers will appreciate the financial pressures created by the care of a ventilated child at home.

Financial pressures also affected families of disabled children generally whilst their child was in hospital (Darbyshire, 1996; Shipley, 1997). The literature about children who are dependent on ventilators, however, tended to concentrate on the issue only at discharge. Some of the United States commentaries seemed to attach little significance to pressures on family finance (Lynch, 1990; Richardson *et al.* 1992; Copeland and Clements, 1993).

The impact and costs of home care on parents

Leonard *et al.* (1993) documented and analysed the economic, emotional and temporal costs on parents of caring for ventilator-dependent children at home in the United States. Leonard *et al.* acknowledged that the lower health service costs and improved health outcomes associated with home care had in fact been achieved at the cost of parents.

In studies focussing specifically on ventilator-dependency, there is evidence that mothers' involvement with their ventilator-dependent children became all consuming of their time (Brust *et al.* 1992; Wilson *et al.* 1998); employment opportunities reduced or ceased (Murphy, 1997; Kagan *et al.* 1998); the burden of providing care increased with the age of the child; and when ventilator users reached adulthood their caregivers voiced most dissatisfaction, resentment and unhappiness (Quint *et al.* 1990; Felinas *et al.* 1998).

Factors that influenced the qualitative empirical study

Review of the literature undertaken prior to the study found that few (if any) qualitative studies around the costs of caring and satisfaction with services had been carried out with children and young people who were dependent on ventilators and their families in the UK. This lack of qualitative evidence around the exploration of costs and satisfaction with services represented a significant

gap in the evidence base and warranted further investigation. In consultation with the advisory group set up to guide this part of the research plan, it was decided that it would be a priority to undertake some qualitative research with children and their parents and offer some recommendations for improving future qualitative research activities designed to explore costs and satisfaction with services in this group of children. The next section will report the qualitative study undertaken subsequently.

7.2 Qualitative study exploring and explaining the costs of caring for children dependent on ventilators and satisfaction with services

Selected findings around costs and satisfaction with services derived from a qualitative study involving 53 children and young people dependent on ventilators and their families are presented here.

Aim

The aim of this aspect of the qualitative study was to describe individual and family costs and levels of satisfaction with services in order to address the following research questions:

1. What are the costs to families of caring for ventilator-dependent children?
And
2. How satisfied are parents and children with the services they receive?

The methods, ethics, sample, data collection and analysis have been described previously in Chapter 4.

Exploratory fieldwork

In addition, findings derived from exploratory fieldwork with professionals are included to supplement the views of parents and children. The methods concerning exploratory fieldwork were described previously in Chapter 2.

Findings

Data reported here are limited to key themes and sub-themes around the children's, their parents' and professionals' constructs concerning the costs of caring and satisfaction with services.

Findings provide a rich description of the financial, emotional and physical burden of care experienced by parents and the time costs involved in being a parent carer. Parents and children also described the inefficiencies and wastage of public resources within and across sectors that added significantly to their overall burden and ability to cope.

Parents and children with 'simple' needs around ventilator-dependency experienced fewer additional costs and higher levels of satisfaction. In contrast, children and young people with 'complex' needs around ventilator-dependency needed more resources and services, and were generally more dissatisfied. The latter families generally perceived the burden of care to be manageable only when a package of support services was in place and worked efficiently. For around half of these families services were frequently found to be ineffective, of poor quality and poorly coordinated across sectors. As a consequence families felt that the financial, emotional and physical burden of care and the time costs involved were excessive.

Key themes around the costs of caring and satisfaction with services

Four key themes around the costs of caring were derived from interviews with parents, children and young people, and exploratory fieldwork with professionals, including:

1. The financial and emotional consequences of prolonged hospitalisation;
2. The financial and emotional impact of living at home with a ventilator-dependent child with complex needs;
3. Social issues concerning 'looked after' children, and
4. The lack of auditing or outcome measures to monitor the effectiveness or appropriateness of the services provided.

Findings from each of the 4 key themes around the costs of caring and satisfaction with services will be described in the following sections.

Key theme 1: The financial and emotional consequences of prolonged hospitalisation

The perceived costs of hospital versus home care

Of the 53 children and young people in the sample:

- 9 children and young people with complex needs were awaiting discharge from hospital at the time of interview;
- 37 children with complex needs were living at home having spent significant periods of time in hospital awaiting discharge, and
- 7 children and young people with less complex needs, used a simple ventilator mainly at night, and only needed a short hospital admission of a few days to get used to the machine and its operation before being discharged home.

The latter group of 7 children mostly did not require employed carers and all appeared to be satisfied with their care and treatment.

The families of 46 children who spent prolonged periods of time in hospital after the initiation of ventilation were completely dissatisfied that their children had to wait months and in some cases years to be discharged home. They thought it was a perverse accounting decision that children with the most complex needs and greatest level of dependency on the ventilator lived in expensive hospital beds when they no longer had a medical need or wanted to be there.

Views and experiences described here are mainly from the 46 children and their parents who spent prolonged periods of time in hospital following the initiation of ventilation. In Chapter 4, prolonged and unnecessary hospitalisation was shown also to have a profound and negative impact on the health and quality of life of children and their families. In this section, the economic consequences of prolonged hospitalisation for the child and their family are explored.

Dissatisfaction around the unnecessary cost of hospitalisation was also a constant theme raised by the young people. Jason, who was a teenager, had sat his maths GCE in hospital. As a part of his course work he had calculated the amount of money the NHS wasted by keeping him in an intensive care bed when he no longer needed to be there. He calculated that this amounted to hundreds of thousands of pounds each year and he could not understand why the NHS would want to waste so much money as well as making his and his family's life a misery.

During exploratory fieldwork, 2 commissioners provided an explanation as to why some accounting decisions appeared to parents to be perverse and not in the best interests of children. They explained that children's intensive care services were usually planned at a regional level and delivered via a 'block' contract with specialist children's services at a regional hospital or NHS Trust. When a child dependent on a ventilator occupied an intensive care bed, other critically sick children were prevented from accessing the intensive care facility. However, when the ventilator-dependent child was discharged home with an expensive package of care, the vacated intensive care bed was immediately filled, thereby increasing the costs to the commissioners of children's services who now had to fund the intensive care bed and the package of care for the

child dependent on a ventilator to live at home. They had seen big increases in the numbers of children dependent on medical technology requiring expensive packages of care in order to live at home. Their experiences matched those of the doctors (described in Chapter 2) who also said that they had seen big increases in the numbers of children and young people dependent on ventilators.

Barriers to discharge and financial consequences

Parents and children identified 5 main barriers to discharge that contributed to their overall dissatisfaction with 'the system': the attitudes of professionals; the lack of joint commissioning of services and accounting responsibility; inefficient discharge management; the cost of, and time taken to, modify or move house; and the slow process of recruiting and training employed carers.

Inappropriate attitudes of professionals

A number of parents felt that professionals used a variety of tactics to keep their children in hospital for months and in some cases years longer than necessary. Several parents said that the possibility of going home was not raised with them for many months despite their children being medically stable and getting increasingly bored and fed up of being in hospital.

The young people used words such as '**stuck**' and '**trapped**' to describe their lives in hospital. They did not feel that they had a voice or had been listened to regarding their wish to live at home. Parents said that the reasons attributed to their child's continued hospitalisation varied, from professionals who considered that they would not cope at home, to those who felt that their child would be safer in hospital. Other professionals did not appear to have the appropriate skills to '**pull the right levers to get things going**' and intensive care staff in particular did not have the time or consider the role of discharge coordinator as their responsibility.

Two families were told that there was no possibility of their children being discharged home on long-term ventilation and they would be likely to stay in hospital indefinitely. Both families rejected this prospect and in one case the

parents asked to be transferred to a hospital that could discharge their child home. In the other case Lisa's parents started the process of discharging themselves. Lisa's mother recalled:

'She was staying in hospital for the rest of her life as far as they were concerned. They were not interested you know, they wouldn't supply us with a ventilator or anything to go home with. So we were like begging the community and big firms (for money).'

Both these children are now thriving at home. The cartoonist associated with the study worked with the children to depict their views and experiences. A common image described was that of a child rubber stamped with ***'NHS property'*** and with no voice (Illustration 7.1).

Illustration 7.1. Young people felt they had no voice whilst in hospital and did not want to be there.



For many families the months of indecision meant that the other arrangements enabling their children to be discharged home had not been put into place. When it was finally agreed that the child could be discharged, there followed many more months and in some cases years of inactivity whilst issues such as funding were sorted out.

During exploratory fieldwork nurses and social workers frequently expressed their frustration at their lack of time to make the arrangements to discharge children home. Seemingly insurmountable barriers such as the lack of money to purchase a package of services and the long process required to provide suitable housing for the child to be discharged were cited. Some parents were also described as having unreasonable expectations and refused the package of support services offered as they perceived it to be inadequate.

The lack of joint commissioning of services and accounting responsibility

The fundamental barrier to discharge, highlighted by all parents who experienced delays, was the lack of any obvious joint commissioning between and within health and social services and the lack of clarity this caused regarding responsibility for funding of the care and services required by the child to live at home. Major delays were experienced because competing stakeholders could not agree funding. This situation was said to be compounded by on-going restructuring of the NHS, such as the amalgamation of NHS Trusts, the setting up of Primary Care Trusts and the reconfiguration of services.

Inequity in the provision of care packages

A lot of dissatisfaction was expressed about access to services; parents felt that disputes over funding resulted in inequity in the provision of care packages. ***'They don't seem to assess your child's individual needs'*** was a common response from parents. They complained that the system ***'wasn't fair'*** and they had little involvement in deciding ***'what they did and did not need'***. There appeared to be much conflict and tension between agencies and individual professionals, and the young people described being caught in the middle.

One parent summed up typical sentiments:

'Most professionals were not acting on the child's behalf. They were defending their own little corners and their own little budgets.'

Children with similar needs appeared to have care packages with big differences in the level of care and services provided. Parents suggested that this was due to a number of factors, including:

- Refusal by authorities to pay for the care package recommended;
- Disagreements over who should pay for carers and services;
- The budget was overspent therefore services were not provided;
- Services were not available in locality;
- Difficulties in recruiting and retaining carers;
- Some parents were expected to act as unpaid carers;
- Parents' ability to fight for services on their child's behalf, and
- Involvement of the media and Member of Parliament to fight for resources.

Many parents had also engaged in protracted disputes over the amount and type of care provided. Some parents insisted on having highly qualified children's nurses, which some funding authorities refused to pay for when less highly qualified nurses or unqualified carers would be far cheaper. Highly qualified nurses were also very difficult to recruit to work in domestic settings whereas employers said that they could guarantee a good response to advertisements in the local media for unqualified carers. There were also prolonged disputes about the amount of care provided. Some families had refused to take their children home until they had been granted what they considered to be an adequate amount of employed care. One mother said that she ***'would not take her child home unless she had 24 hour care'***. This had resulted in her child staying much longer in hospital until a compromise could be negotiated.

A contrasting point of view was expressed by one NHS manager of community services. She said that it was important that families experienced a short period of time in their home each week when they could be together as a family without the intrusion of employed carers. This allowed mums and dads to parent their children and spend time together in privacy. In her experience, some

parents tended to rely on employed carers and took a peripheral role in the life of the child dependent on a ventilator.

One NHS community manager also said that although some parents insisted on having qualified children's intensive care nurses, it was difficult to retain this type of nurse in the community. Exit interviews with nurses had revealed that they quickly lost their intensive care skills and nurses missed the working environment of the intensive care unit. Most left within 6-12 months and returned to work in a hospital environment; the manager considered this a waste of money in terms of investment in recruitment and training. There was also a national shortage of children's intensive care nurses and the manager considered that it was a waste of their expensive training to deploy these highly qualified nurses in the community.

During exploratory fieldwork, 2 commissioners of services said that most finance directors had an obligation not to overspend and their performance-related pay was dependent on the financial position of their organisation. There was no clear guidance as to who should pay for what. They explained that at a political level, the Government had yet to establish an activity-based costing system that determined the accounting responsibilities of the various funding agencies for the substantial cost drivers (such as the cost of a ventilator or the funding of full-time carers).

One commissioner gave the example of a child that required a specialist chair. The need for the chair had been identified by the school, but the education authority were refusing to pay for it as seating was considered to be the responsibility of the social services department. In turn, the social services department were overspent and refused to pay for a chair that was going to be used predominantly within the school.

On a similar vein, much tension was apparent when an assessment of need had been made by a specialist hospital outside of the region where the child lived. This happened frequently as many of the children and young people said they had been transferred to specialist hospitals many miles from their home.

The local Primary Care Trust, health authority and social services department had little input into undertaking this assessment and yet were responsible for funding the resultant package of care recommended by the specialist centre. One commissioner said that these assessments were akin to wish lists and raised unfairly the expectations of the children and their families, especially when there was insufficient funding to pay for the package of services recommended.

Fund raising and charities

For some families the shortfall in NHS and social services budgets was made up by fund raising and involving charities. A few parents said they had purchased and maintained expensive and essential equipment ranging from ventilators to wheelchairs. They described using the media to highlight their child's plight as ***'a humiliating experience'*** and ***'an invasion of privacy'***.

Independent brokerage

Kathy, whose child had been in hospital since birth and was now 3 years old said that she was ***'locked in dispute'*** with her hospital consultant. Kathy was fed up with what she considered to be the disorganisation within the state provision and wanted to investigate the system of 'independent brokerage' whereby a fixed sum of money is allocated to the parents to purchase services for their child. Her consultant had never heard of this system of payment before. Whilst the protracted dispute over independent brokerage was unresolved her child remained in hospital unnecessarily.

When questioned, none of the other families who used state provision had been offered independent brokerage schemes. In contrast, one family who had secured a financial payment for their child through litigation, purchased services for their child and said they liked having choice and financial control over their child's care package.

Inefficient discharge management

Parents complained that NHS and social work staff appeared to have little previous experience or guidance to draw upon and in most cases no one

person was identified as having responsibility for organising their child's discharge home. Communication between the discharging hospital and other hospitals, social services departments, education authorities, the voluntary sector, and especially with parents, was said to be poor and infrequent.

Having complex needs and being in a hospital a long way from home was said by parents to make the discharge process far more complicated. Parents complained that **'things didn't happen quickly'**. They talked about the stress this caused them and the actions they were compelled to take. Most parents had to **'phone everyone constantly to keep things moving'**. Parents consistently said that the problem was **'nobody really knew what they were doing'** and **'it was all down to poor communication'**.

Jason's father outlined a common view:

'The problem was nobody really knew what they were doing because nobody had experience of tetraplegics and ventilator-dependency. They didn't really know what was expected and what was feasible.'

Most families despaired at how slowly things were organised and how little control they had over events. Jack's mother who was dissatisfied with how things were progressing with her son's discharge said:

'The local health authority were blaming the specialist centre and the specialist centre were blaming the health authority. It was like a game of ping pong.'

These comments involving blame and counter-blame were typical of many conversations with parents and the young people. Families were left feeling angry, helpless and bewildered.

Discharge-planning meetings should ideally be organised at appropriate intervals so that the relevant people involved can meet and review progress. In reality, the young people and especially their parents were very dissatisfied with

the overall organisation and management of meetings. The reasons given for this are summarised in Figure 7.1.

Figure 7.1. Problems parents encountered with discharge-planning meetings.

- No discharge-planning meetings were arranged;
- Too many discharge-planning meetings were arranged;
- Parents/young people were not invited or not informed when meetings were scheduled;
- Parents travelled long distances to attend;
- Meetings were frequently cancelled without informing parents;
- Parents were excluded from all or part of the meeting;
- Professionals who chaired the meetings were inconsistent, ineffective and did not introduce parents or involve them in the discussion;
- Too many people were present at meetings;
- Key people frequently sent apologies or never attended meetings;
- Arguments and heated discussions occurred between professionals (usually about funding or whether the child should go home);
- Things were agreed and no action taken;
- Nothing was agreed, and
- Minutes were not recorded or not given to parents.

Parents were angry and frustrated at the way they had been treated. One parent recounted:

'We were not allowed into any meetings until the last bit and then we were sent out again while they discussed us.'

She went on to say:

'Before we were due to go home they had one big meeting at the hospital. There were about 25 people coming in, including the cleaners nearly. We were only in for 5 minutes before we were sent out again.'

Another parent recalled:

'We had meetings and meetings and meetings that got nowhere. social services live to hold meetings and produce paper.'

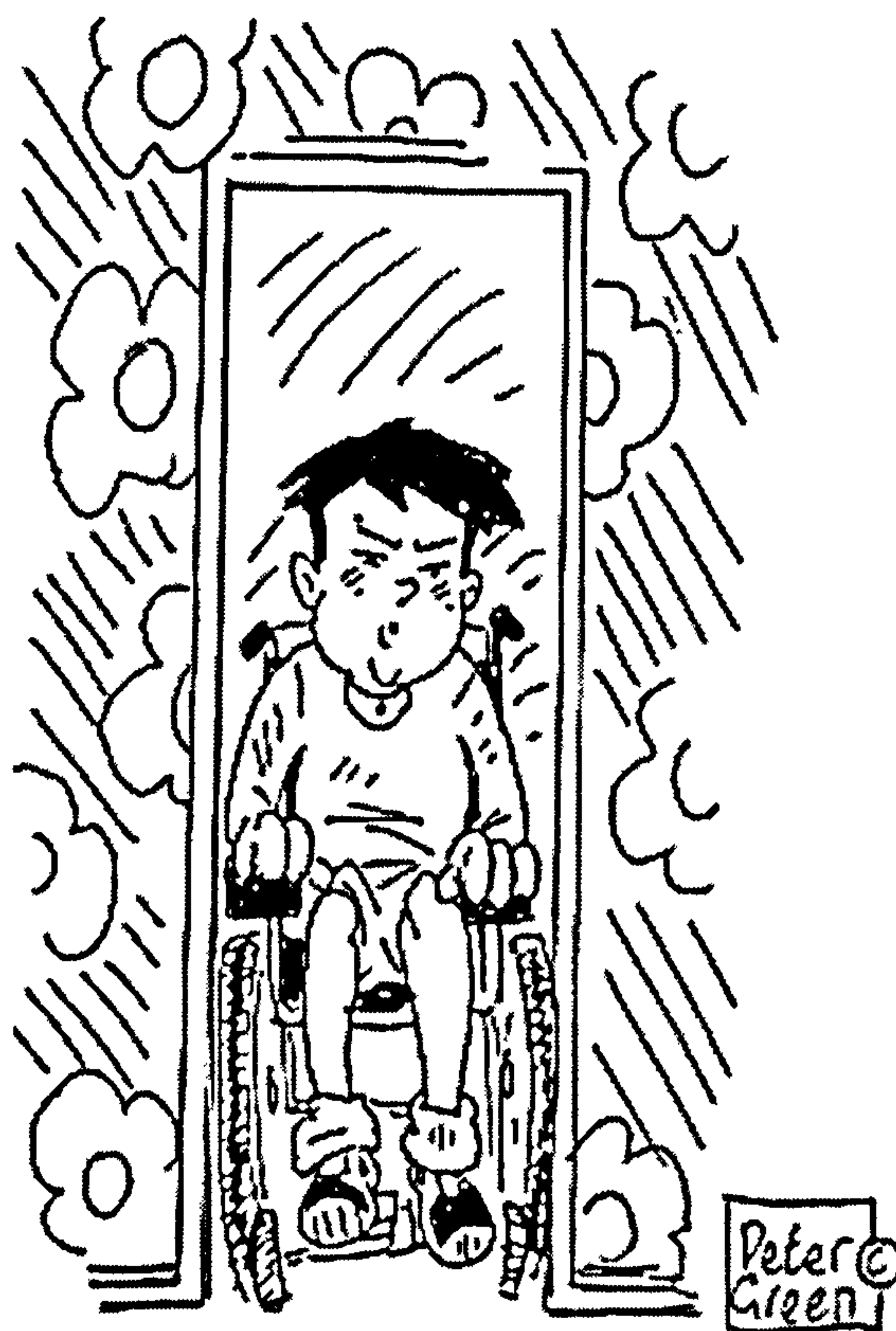
During exploratory fieldwork a discharge-planning meeting took place, which involved 22 professionals and 2 parents; many of the people had travelled over 60 miles to attend the meeting that went on for 3 hours. The staff and travel costs incurred were substantial as most of the professionals would have allocated an entire day in their diary in order to allow sufficient travelling time to attend. One parent who was self-employed said that they had taken the day off work and had lost out on vital earnings.

During a later interview with the parents involved, the mother said that no one person was responsible for coordinating actions across health, social and education services and little progress appeared to have happened since the first meeting 3 months earlier. It had taken a further 2 such meetings before a discharge date had been negotiated and in her opinion the entire process was disorganised and a waste of public money. She felt that she had taken on the role of organising her son's discharge and had run up large bills on her mobile phone as she had to phone professionals and agencies from the hospital where she was living with her son.

The cost of, and time taken to, modify or move house

Many families found that their home was unsuitable for their child's needs and expensive adaptations were required in order for the child to be discharged home. The suitability of a child's home was also highlighted in Chapter 4 as having a major impact on the child's quality of life. The cartoonist associated with the study depicted a common experience related by children and young people who used wheelchairs and said that they could not get through doorways (Illustration 7.2). Parents said that it was a big expense to have doorways widened and most found subsequently that corridors and the small size of rooms in ordinary domestic housing stock was still inadequate for a child to manoeuvre a wheelchair with ease.

Illustration 7.2. Children and young people found that their homes did not always meet their needs and ordinary houses were particularly unsuitable for children who used wheelchairs.



Although most families had to have some building work and alterations undertaken on their homes, only 6 children remained in hospital for a substantial period of time whilst the work was being carried out. For the remaining families the very long wait for funding agencies to agree to pay for a package of care and recruit a team of carers was far longer than the period of time taken to complete the building work and alterations they had paid to have on their homes. The experiences of Mathew's father were typical of many families. Mathew (age 5) had become dependent on a ventilator following an accident. His parents owned their own home and had re-mortgaged their house to pay for a large multi-room extension for Jack and his care team. Mathew's father explained:

'The extension for Jack was finished months ago. It is all kitted out and just sitting there empty. The hospital have not even started the process of recruiting carers. In the meantime, Jack lives in an intensive care unit which is awful. The stupid thing is that they send him home in a taxi

for the day whenever they have a free nurse, but he has to return to the hospital when the nurse's shift ends..... He even goes to a local school up the road during term time whilst being an intensive care patient!

For one child whose discharge was significantly delayed by housing problems, her family had to be first re-housed by the local authority and then substantial alterations had to be undertaken to the property. The involvement of the local housing department added yet another level of complexity to the discharge procedure. The child's mother reported that her daughter spent many months in an expensive hospital bed because of a general lack of communication and overall inefficiency within the housing department concerning the completion of a number of relatively inexpensive and yet essential jobs, such as putting in additional power sockets.

Parents found themselves having to unexpectedly increase mortgages and take on additional bank loans when they least expected it. They felt that advice given was sometimes overwhelming. Professionals used persuasive language such as ***'you will have to do this with your house, you will have to have one of these'***.

Other families considered their current homes could not be adapted and started the process of looking for another property, or a plot of land to build a purpose designed house. This was said to be a very time consuming and stressful process.

The biggest problem identified by parents was the inequity of the disabled facilities grant, which was supposed to help make homes accessible and safe for their disabled children to live in. This benefit was means tested and parents felt that the calculations were ***'unfair'*** and ***'unjust'***. Parents said that their incomes were means tested and not the income of the child as was the case with disabled adults. The means test did not include actual expenditure or account for the additional costs of caring for a disabled child.

Applying for the disabled facilities grant was said by parents to be bureaucratic and there were insufficient occupational therapists to help with assessments. Parents said that they had little choice about the design of additional floor space and despite the fact that parents with higher incomes made a major financial contribution, the tender was awarded by the local authority housing department to an approved builder. In particular, the disabled facilities grant could not be used to pay for an additional kitchen and sitting room for carers even though privacy and space for carers was highlighted in some cases as absolutely essential by both parents and occupational therapists.

Another problem that hampered progress towards discharge was the conflicting advice received from various agencies. Jason's parents were planning to have a lift installed in their home and a firm of architects had drawn up plans, which needed to be approved by the borough engineer. His dad recalled a conversation with the borough engineer during a site inspection of their home:

***Dad: 'I asked: Can you get the wheelchair up in the lift?
And the engineer said 'No I wouldn't have thought so, it will
be too heavy, too long'.***

***So suddenly all of the advice we were getting from social
services, occupational therapists, engineers etc about
adaptations to the house didn't match.'***

Jason's parents wasted vital resources on having these plans drawn up unnecessarily, as the borough engineer recommended that they built a ground floor extension for Jason that did not require a lift.

Sarah's mother explained that her local housing association had installed an expensive electricity generator at their home and she was not sure why a generator was required when the provision of mobile battery packs would have been more practical in the event of a power cut and far cheaper. The generator had also hampered her wish to move house as it was unlikely to be required by another resident.

Once home, families noted that some items of equipment were never used or totally unsuitable. Other families did not have all the things they required to care safely for their child. Jessica's father said they went home with very little back up equipment and Lisa was not initially provided with an essential oxygen monitor for home use. She also had to manage with large oxygen cylinders when an oxygen concentrator may have been easier, more convenient and less expensive.

Sanjay aged 12 also explained that his family had received bad advice concerning the extension to their family house. Sanjay who was tetraplegic and dependent on a ventilator 24 hours per day lived in hospital for over 5 years whilst his family house was extended with the aid of a disabled facilities grant. Within 3 years of being discharged, Sanjay had grown considerably and could no longer get into his purpose designed bathroom as the space was too small to manoeuvre his new larger hoist; this meant that his carers now had to give him a bed bath which he hated. On inspection, Sanjay's bathroom was very small and yet there was a large garden outside which would have allowed for a larger extension. In addition, as he reached adolescence, he said that he did not always want his carer to spend so much time in his bedroom. As the disabled facilities grant did not pay for a kitchenette or sitting room for carers, a corner of Sanjay's bedroom had been equipped with a microwave, fridge and chairs for his carers; this arrangement was no longer acceptable to Sanjay. He felt that if his parents could speak English more fluently they would have been able to convey their concerns about the proposed design at an early stage and prevent the local authority and the family from wasting money on an inappropriate design.

Other families had to rely on a housing association or the council to provide adaptations or alternative accommodation. These adaptations were often included in the care package and some families had to wait months for a budget to be agreed. Some councils were very inefficient at getting work completed and this caused a lot of dissatisfaction. One mother said:

'I had to phone up and say the jobs weren't done. They didn't believe us so they came out and had a look!'

Apart from space for the child, the second major issue cited by parents and children was the lack of storage space in their homes for disposable equipment, medical supplies and bulky items of equipment such as standing frames and wheelchairs. In particular, local authority approved extensions had not provided sufficient storage space. The author visited almost every family at home and witnessed at first hand the problems encountered. These issues could have been overcome if the council approved architects had taken into consideration the child's need for equipment and supplies of disposable goods such as boxes of suction catheters, incontinence equipment, ventilator circuits etc. Many parents were contemplating additional building work or moving house to overcome problems with storage and one parent had purchased a mobile caravan to store disposable equipment and supplies, which was parked on the driveway.

Some families went home with minimal adaptations and wanted time to make their own plans for the future. Some of these plans were dependent on legal proceedings that commenced following an accident involving the child. Legal proceedings appeared to take years to complete and 4 families in this study had received either interim or final payments. One child who had been involved in a car accident and who participated in the study was subsequently awarded over £3 million following legal proceedings; his parents planned to use an interim payment to build an architect-designed house on a plot of land.

Many families had experienced a change in circumstances since they were originally assessed for a disabled facilities grant and had adaptations undertaken to their house. One family explained that they had a further baby and their adapted house was now too small. They had applied for a further disabled facilities grant to build an additional bedroom for their expanding family on the grounds that the new baby would have to share a bedroom with their disabled child as the third bedroom was being used by carers. The family had the backing of their local Member of Parliament and had highlighted their situation in the local media. Another family had sold their house soon after

completion of a multi-room extension which was part funded by a disabled facilities grant as the father got a new job in another area. When interviewed, this family were looking to buy another house and wanted to apply for another disabled facilities grant with a different local authority.

A local authority housing officer who was interviewed during exploratory fieldwork provided a different perspective. She said that the grant from central Government for housing adaptations had increased substantially over the last 5 years, but demand for disabled facilities grants had outstripped supply. She supported the means testing of the benefit as in the current housing market an extension added significant value to the property. Open plan living with fewer but bigger rooms and polished wood floors that grants routinely funded for people with wheelchairs had recently emerged as a very popular design concept amongst the general population. She cited the case of one family whose 3 bedroom detached house increased in value by over £60,000 following a £20,000 grant to provide a 2-storey extension with a large bedroom, en-suite bathroom and downstairs open plan living area with polished wood floors. The family subsequently sold the adapted property and purchased a large 5 bedroom house nearby, thereby reaping the benefit of the grant and in the eyes of the housing officer *'ripping off the tax payer'*.

Good Practice

Many families who required re-housing with the local authority often had to wait months as there was an acute shortage of larger properties in almost all areas caused by the 'right to buy' scheme which resulted in the loss of larger properties from council stock. One single mother described that her local authority had purchased a large detached house for her family to move into. She said that she had been kept well informed of progress and this had been a good way of overcoming the shortage of larger council houses.

In recent years there had also been substantial local authority and housing association building programmes. Revised planning regulations had required a percentage of new properties to be suitable for disabled people, and families in this qualitative study benefited by living in such properties.

Despite lengthy arguments about funding for disabled facilities grants having to extend mortgages, and dealing with public sector housing departments or associations, nearly 3 quarters of families said they lived in nice homes and were satisfied with their housing at the time of interview.

The slow process of recruiting and training employed carers

The process of recruiting carers could not begin until funding was agreed, and as discussed previously, in most cases this caused significant delays to discharge and much dissatisfaction. Having agreed funding, the general process of advertising, recruiting and training staff to work in the home was criticised further by parents as being too slow and overly bureaucratic. For example, one Primary Care Trust had a moratorium on the recruitment of staff as their budget was overspent substantially. Some families were told it would take up to 2 years to recruit and train a team of carers. Parents also said that there was a poor response to job advertisements seeking to recruit qualified nurses to work in domestic homes. There were also bureaucratic problems about allowing staff from other organisations and sectors to learn how to care for children within NHS hospitals before discharge.

Three young people who had been assessed as needing employed carers at home were eventually discharged without any. This was due to either failure to agree funding or failure to recruit, train or retain appropriate carers. In 2 cases, the parents both gave up full-time work. In the third case, the parents continued working and were initially able to manage their son's night-time ventilation with the support of their family. Ajaz's father said that he felt his young son would have been in hospital a further 2 years had they waited for the health authority to agree funding to employ the carers they wanted.

During exploratory fieldwork, NHS and social services managers said that they had also experienced problems with the unacceptable attitudes of a small minority of parents. For example, one family demonstrated racist attitudes towards candidates. The managers were worried that they would be subject to legal proceedings and as a result the process of recruiting carers had been very expensive and ineffective. Parents generally needed help to understand their

obligations concerning equal opportunities and employment law and managers were worried that ethnic minority nurses may be subject to inappropriate attitudes in the home.

The additional costs incurred by the family whilst their children were in hospital

All parents said that having a child in hospital for prolonged periods of time was a financial burden as they had to:

- Drive long distances to the hospital;
- Make frequent journeys to the hospital;
- Pay for car parking;
- Purchase hospital food;
- Pay for accommodation to stay near their child;
- Pay for additional child minding, and
- Reduce or give up paid employment.

Parents said it was difficult to juggle their responsibilities and they found hospitals to be very inflexible. Lack of support and absence of family friendly policies such as allowing siblings to stay at the hospital caused additional financial and emotional stress. Some parents arranged for other children to stay with grandparents, relatives, friends and paid child minders. Other families managed a rota system of people to care for their remaining children at home. Parents undertook an enormous amount of organising to keep these arrangements going for months and in some cases years. Mothers talked of being separated from their husbands and other children and passing like ships in the night as they swapped roles at predetermined times. For example, Sarah's mother organised her life for over 6 years around one daughter at home and one daughter who was living at the local hospital. This meant that she spent a lot of money on petrol, car parking and extra child minding. Although Sarah's mother did receive reimbursement for the costs of travel to and from hospital, she felt that she was still worse off financially than if her child was living at home. If her child were living at home she would be entitled to claim additional welfare benefits (such as the disability living allowance), which were currently denied as she lived in hospital.

Some parents felt that they had to go to *'extraordinary lengths'* in order to remain near their children. Attitudes varied widely from hospitals that encouraged parents to stay with their child free of charge to hospitals that did not provide any parental accommodation. Five parents talked of having to *'find and rent accommodation'* locally in order to be near their children. Charities and volunteers based at these hospitals helped parents find somewhere to stay. These parents talked about the financial burden of having to support the family home and rent additional accommodation at a time when they were coping with an emotional crisis and substantial loss of income when one or more parents could no longer work. One mother said that she was forced to leave her family home and rent accommodation near to the specialist hospital where her son was being treated after an accident. To help pay for the extra costs she got a part-time job in a local chain store and visited her son whenever she could. At weekends, her husband travelled from their family home 100 miles away and she went home for the weekend to see their other children. This routine continued for 3 years until their son was discharged home. This mother calculated that it had cost them *'thousands and thousands of pounds in additional rent and travelling expenses'*. Another father said that his wife had driven 32 miles per day and spent £2 per day to park in a hospital car park for over 3 years; he calculated that the car park charges alone amounted to over £2,000. All parents whose children spent prolonged periods of time in hospital said that much of the financial burden experienced by their family was unnecessary, as their children could have been discharged home far sooner thereby reducing their costs.

Loss of or reduced employment for one or both parents

Almost all parents said that they faced loss of, or reduced, employment as a direct result of their child's hospitalisation and subsequent need for long-term care. Loss of income was particularly acute for self-employed and single parents who were sole breadwinners. One couple who ran a successful business said that they had virtually stopped trading since their child was critically injured in an accident. Mothers in particular said that their ability to remain in paid employment had been severely affected; most said that they were unreliable employees as their child's needs always had to take

precedence. Almost all children and young people required long-term care and in most cases parent(s), primarily mothers, were either required to give up work or be the sole or additional carer for their child.

One father also said (off the record) he did not intend to go back to work until the disabled facilities grant had been sorted out as he would have to make a far higher contribution if his income was taken into consideration.

Parents as unpaid labour

All of the parents had to learn how to care for their children's complex needs. Being resident in hospital provided them with every opportunity to gain competence and confidence in a relaxed environment. Parents spoke of being **'involved'** and **'learning'** about their child's care. However, there came a point at which most parents said they were used as unpaid labour and were expected to care for their children in hospital; some parents said that this was for 24 hours a day. Almost all parents described staff shortages and having to be around to make sure that their child's needs were met. Some parents were very resentful of this perceived obligation to provide an unlimited supply of unpaid labour.

Good Practice

Few examples of good practice regarding the discharge of children with the most complex needs were evident. Only Mary's mother was relatively happy with her daughter's discharge home from hospital. She put this down to:

'Mary being discharged at the beginning of the financial year when the health authority still had money in the pot.'

Other families felt that eventually they got much of what they wanted but:

'Had to fight every step of the way.'

Parents were more satisfied with those hospitals that had recently employed discharge coordinators who managed all aspects of the discharge process and who liaised with all agencies and sectors involved.

Key theme 2: The financial and emotional impact of living at home with a ventilator-dependent child

Reduced earning capacity and reliance on welfare benefits

Once home, families continued to experience reduced opportunities for employment and this had a major impact on their ability to earn money. Parents said that their child's need for adapted housing and specialist services reduced their mobility in terms of employment and limited opportunities for promotion. Mothers who took an active caring role said that their opportunities for employment were limited as they could be unreliable and require frequent absences because of their child's illnesses.

The level and amount of employed care could also limit the employment opportunities for parents. For example, some parents were required to cover a number of shifts per week and unqualified employed carers required constant supervision by parents at all times. In contrast, qualified nurses could be left unsupervised and this enabled parents to engage in employment outside of the home.

Of the 50 children whose parents were responsible solely for their welfare, 20 households had no adult in employment and were reliant on welfare benefits. For most of these twenty households, their main income was linked to their disabled child.

It was also evident from talking to parents that many experienced high levels of stress and several admitted to taking anti-depressant medication to help them cope. A typical scenario was that of George who explained that he had a serious accident as a teenager and his dad suffered a nervous breakdown soon afterwards and was now taking Prozac. The anti-depressant medication had improved his mental state but he was unable to return to work.

Family breakdown and financial hardship

Fifteen children (28%) had experienced the divorce or separation of their parents. Mothers said that the breakdown of their marriages and relationships had had a considerable impact on their finances. A typical example was that of

Julie age 11, who had complex problems, and her family. Her mum explained that Julie's dad had left the family soon after her birth as he could not cope with her disability. He had since moved away and remarried but despite being a high wage earner had not contributed any financial support to the family or maintained contact with any of his 4 children. Julie's mum was not able to work as she cared for her daughter; as a result the family relied on welfare benefits as their main source of income.

Inefficient on-going management of care packages

Significant problems were described by around half of the families in relation to the ongoing management of care packages that were delivered by staff from various agencies and sectors. Some families were told they were not entitled to look at or have a copy of the document outlining their care package; this was felt to be unnecessary bureaucracy. In contrast, other families said that they had entered into a signed agreement with their newly formed Primary Care Trusts that outlined explicitly the care and services they were meant to receive.

Parents described a number of problems, including: lack of coordination of services; no clear role description for carers; lack of protocols and guidelines (for example, health and safety); retention of carers; increased household costs and inflexible care packages that did not respond to their changing needs.

Lack of coordination of services

Delivering complex packages of care to children and young people at home is a relatively new concept and parents had experienced a lot of problems with having to deal with so many people and agencies. In Chapter 4, parents said that too many professionals had become involved with their families and this had had a negative impact on their quality of life. Around half of mothers said that they took on the role of coordinating all the various professionals, teams, agencies and hospitals. One mother explained that she rang 32 different people on a regular basis and incurred large phone bills as a result.

Only half of the families could identify a key worker who was in a position of sufficient authority to liaise between different teams and sectors. Many parents

had also turned to the voluntary sector to provide additional services on top of their agreed state-funded care package. For example, one mother had arranged for Barnardos to provide an extra carer to help on schooldays and the charity Crossroads provided additional respite care at weekends. This provided the support that she required, but resulted in a fragmented package of care that was left largely to the mother to coordinate.

During an interview with a NHS manager of community services, it became apparent that she rarely had a clear picture of the services each family received, as in her experience families sought additional services from the voluntary sector but were afraid that if the NHS manager found out, the package provided by the NHS would be cut. This was confirmed by Jacob's mother who had an additional carer from the voluntary sector that she did not really need but was afraid that if she gave up these hours she would not get them reinstated if her child's needs changed.

Review of care packages and the appropriate use of staff

Thirty children currently had employed carers at home and 9 children were in the process of recruiting employed carers in order to be discharged home. Managers undertook reviews of the activities undertaken by carers in the home and regularly found that carers were under-utilised or used inappropriately. One example of under-utilisation was a mother who wanted to do everything for her child, leaving the carer with little to do all day, which was viewed as a waste of resources by the NHS manager.

Having spent prolonged periods of time in hospital, parents had got used to having awake carers at night to look after their children. When the children were discharged home the tradition of having an awake carer continued in most cases. However, time and motion studies undertaken by managers had revealed that many night carers were very under-utilised and had little to do once the child was asleep; some carers were watching television all night. During exploratory fieldwork, managers frequently said that they tried to replace qualified nurses when they left with untrained carers as they tended to stay in

post longer, were less likely to exert their opinions on the family, and were cheaper to employ.

Parents said that nurse managers were always keen to reduce the amount of nursing hours that their child received and they did not like the pressure that this put them under. On occasions reductions in carers' hours had been done without consultation with parents who were informed of changes to their care package by letter; this was thought unacceptable and not in the spirit of working in partnership.

For both parents and carers there was a lack of clarity regarding roles and responsibilities. Four mothers complained that nurses and trained carers, who were used to being employed in hospital, did not consider domestic cleaning to be part of their job description. In hospitals, cleaners were employed to clean equipment, bathrooms and areas used by patients. These mothers said that they believed that the carers should clean up after themselves, or a cleaner should be employed to clean the mess created by carers in their homes.

Many carers reported that they performed inappropriate tasks; some by choice, others by request. One carer who sat by a child's bed in a darkened room all night said that she was bored and found it difficult to stay awake. She had asked the family if she could do their ironing and other domestic chores at night in order to remain stimulated. Her NHS manager felt that these activities were inappropriate and arranged for the carer to undertake a distance-learning course at night.

Some care packages also included flexible additional hours of carer support so that parent(s) could go out in the evening or attend an evening class. Mary's mother who was single had asked for additional hours but the carer had reported back that the mother preferred to spend time with the carer in her home as opposed to going out of the house as planned. During this time, Mary's mum also wanted to attend to Mary if she needed anything. The NHS Manager said that she could not justify providing a carer to spend an evening at home chatting to the mother.

During fieldwork, the author also observed employed care teams at work in the family home. Most of the employed carers were seen to be busy and appeared to enjoy engaging with the child and their family. In 4 families both parents were unemployed and a full-time team of carers had been employed to care for their child. In one family, a 10 year old child was looked after simultaneously by 2 parents, one carer and a trainee carer. When the child was interviewed she said that she wanted *'more time on her own'* and *'hated being the centre of so much attention'*.

Parents as carers

There was a lot of resentment and dissatisfaction expressed by some, but not all, parents concerning the expectations placed on them by NHS and social services managers. A common issue was the expectation that one parent would be available as 'back-up' to the unqualified carer in their home at all times. This 'back-up' arrangement was required either because unqualified carers needed supervision whereas qualified nurses could work unsupervised, or because two people were required to lift and reposition the child. In a few cases, the children were relatively unstable and two people were required in case of an emergency.

In addition, almost all parents said that sickness and absence of carers was a major problem and during such episodes parents were required to cope on their own at very short notice. Almost all parents provided care for their children without additional financial reimbursement other than statutory welfare benefits such as the Carer's Allowance. One mother however felt that the Carer's Allowance was insufficient recompense for caring for her child. She had insisted that she should be paid as a carer for working shifts with her child. The child's grandmother and several other relatives were also employed by the NHS as unqualified carers for the child.

Retention of carers

Almost all parents who had employed carers in their home experienced a high turnover of staff, especially amongst trained nurses. Frequent turnover of staff made parents feel very dissatisfied, but they did not know how the situation could be rectified. One nurse manager explained that younger nurses were

happy to gain 6 months of experience working in the community and then wanted to return to a hospital setting or gain more experience elsewhere. Some nurses and carers found that they could not cope with the demands of working within a family home for long periods of time.

Some carers were described as incompetent or incompatible and parents were happy to see them leave; others were considered invaluable and children in particular were heart broken when they left.

Good Practice

Just under half of the 44 families whose child lived at home said that things were working well and they were reasonably satisfied with their care package. Parents and children identified carers and professionals that had made a real difference to their lives. Parents were particularly happy when they worked in partnership with a key person who had the authority to make things happen and who was able to respond quickly when things needed sorting out. The success of the key working partnership also depended on the compatibility of parents and professionals.

Nurse managers were looking for innovative ways of retaining carers, such as rotating them around different families in the same area and offering them additional experience in hospital. Support groups had been set up where carers could meet to discuss the pressures of working in domestic homes and share solutions.

Equipment and disposable supplies

Parents expressed concern at the amount of money the NHS wasted unnecessarily on equipment and disposable supplies. For example, Jessica's father was surprised to find their health authority opted to lease equipment instead of buying. He calculated that this was a far more expensive option:

'They leased the ventilator for the first 4 years. They are still leasing the carbon dioxide monitor, I worked it out they

was paying £85,000 for equipment.’ (£85,000 to lease a ventilator that cost less than £10,000 to buy)

He went onto say:

‘ We kept telling them it was a waste of money.’

A new business manager at the health authority eventually listened to the family, confirmed the facts and immediately rectified this unnecessary waste of money.

Servicing of equipment

Families found themselves having to arrange regular servicing of equipment. Some items were covered by a maintenance contract and routinely serviced by the company who provided it. Many parents did not have a copy of this contract and were unaware of the level of cover they had or who to contact. Some parents however said they had a named contact at the local hospital who would provide replacement parts or equipment as requested. Many items of equipment had been purchased by private funds and parents were not clear who (if anyone) was responsible for maintaining the equipment. Some items of equipment that were purchased privately did not appear to have ever been serviced, which is a potential safety hazard.

The issue of safety and the use of medical devices in the home was raised in an interview with a representative of the Medical Devices Agency. She said that there were serious risk management issues concerning the issuing of hazard warning notices as the Medical Devices Agency had no idea who to contact when privately purchased equipment was used in domestic settings. One example of this was a new type of ventilator that was rolled out into the community and an internal part was subsequently found to be faulty. A hazard warning notice was issued by the Medical Devices Agency but concern remained as most of these ventilators were used in domestic settings and the manufacturer did not have a record of their precise location.

Distribution of disposable supplies

Managing technology-dependent children and young people in the community had created a need for a regular distribution network of disposable equipment and supplies. The distribution of supplies appeared to be working better for some families than others. Some Primary Care Trusts had placed contracts with private companies to supply and deliver disposable equipment and supplies to the house. Thirty-six families talked about the benefits of having supplies delivered to their home on a regular basis. Other parents had to collect large and bulky items from suppliers on a regular basis, which was expensive for parents in terms of transport. Most families obtained items via several routes, which required a lot of organisation. For example, one child obtained items such as oxygen, enteral feed and drugs on prescription from the GP; oxygen was subsequently delivered by lorry direct from the supplier, enteral feed and giving sets were delivered via courier direct from the company and the remaining items were picked up at the local pharmacy; continence supplies were obtained from the specialist health visitor at the local clinic; items such as sterile dressings, surgical tape and suction catheters were obtained via the district nurse; all supplies for the ventilator were obtained from the regional hospital and were either sent by post or picked up by the family. Some mothers were also required to supply schools with disposable equipment for their child.

Some parents had control over ordering their disposable supplies and said they were very careful about not ordering too much at one time. These parents had created their own databases of supplies and had calculated minimum stock levels.

Around two thirds of parents said that they had problems with their current ordering system. One example of waste cited by a mother was the fact that some items such as bottles of electrolyte solution were purchased in bulk by the NHS because of discounts, but the family were unable to use all the bottles before the expiry date. The mother went on to explain that each bottle cost around £50 and she was only able to use around half of the 10 bottles in one bulk pack. She could not understand why one bulk order could not be shared amongst 2 or 3 children in the community.

Many parents said that they needed more information about such things as tracheostomy tubes and appropriate connections. One mother said that she had phoned all the major manufacturers as she was unhappy with her daughter's tracheostomy tubes, which were rubbing her neck.

A small number of parents were also using disposable equipment inappropriately. One mother insisted on changing her child's tracheostomy tube every day and a half, despite the fact that this was not in her child's best interests; local clinical guidelines recommended changing the tube once every 2-3 weeks unless blocked. Each tube-change cost around £50 and her annual bill for tracheostomy tubes alone came to over £12,000. This mother complained that the NHS Manager was *'getting at her'* and felt that the manager was trying to save money and compromise her child's safety; whereas in reality, the mother was putting her child at unnecessary risk of trauma to the trachea (wind pipe).

Another mother said her child destroyed 2 adhesive skin probes per day that were used to measure oxygen levels. These probes wrapped around the big toe and the child who had learning difficulties kicked and twisted his feet until the probes were removed. These probes cost around £20 each and her annual bill for oxygen probes alone came to over £14,500. The mother said that no one had questioned the number that she was ordering or suggested that she might not need to monitor her child's oxygen levels all the time especially as he had a full-time carer to monitor his wellbeing.

Wheelchairs

In Chapter 4 having the right wheelchair was said by children to have a positive impact on their quality of life and ability to be included in social activities. The provision of NHS wheelchairs and buggies was also singled out for particular criticism as the system of supplying and fitting them was bureaucratic, slow and wasted money. Some children had waited around a year from initially being measured for a wheelchair to its eventual delivery. Everyone thought that this was far too long, especially as children were growing in size and the delay

limited the use of the chair in the long run as they were likely to outgrow it without getting maximum use.

Ajaz's story was typical; despite being in hospital for over 2 years he did not receive his own wheelchair before being discharged home. Ajaz's father said:

'There was not a wheelchair that he could come home with ... he had a temporary buggy, but it was so long he could not sit properly.'

Jack (age 8) who lived at home was awaiting delivery of his fifth NHS wheelchair in 5 years and said that his previous wheelchairs had not fitted correctly. Jack said that he was a difficult shape to manage and as he grew his spine was not able to hold his body weight. His mum could not understand why he was not referred to a specialist centre who were experienced in managing children like Jack. One such unit was the rehabilitation engineering unit at Chailey Heritage in Sussex. Jack's mum had made inquiries and found out that a multidisciplinary assessment and adaptation of a wheelchair cost around £2,300 on top of the cost of the wheelchair (around £6,000). Jack's mum felt that this would be money well spent if Jack ended up with a wheelchair that was comfortable and he could use.

Having the wrong wheelchair could also impact negatively on the child's health. Jason who was a teenager and tetraplegic had been waiting for his new wheelchair for months. He said:

'I've been sitting in this crap chair for 4 months, it's doing me no good, I've got spinal curvature now.'

Jason may require surgery in the future to correct this curvature, which may have been avoided if he had been provided with the right body splints and properly fitting wheelchair.

Some parents said that NHS wheelchair services provided equipment through joint partnership arrangements with other services such as the local education

authority. For example, Mark was obtaining a wheelchair from the NHS wheelchair service, but his local education authority was providing adaptations to the chair so that he could access surfaces of different height at school. In practice, Mark said that his mum had to do all the coordination and neither service seemed to communicate effectively. Having to liaise with 2 agencies added a layer of bureaucracy and resulted in additional delays.

Children who received their wheelchairs more than 5 years ago complained that there was little choice in terms of design of NHS wheelchairs and parents said that if they chose to purchase a wheelchair from the private sector then they were saddled with the entire cost. Wheelchairs and buggies obtained directly from commercial suppliers could be delivered far sooner and some parents gave up on the NHS in order to get the equipment they wanted within a reasonable time frame. Fay's mum decided to purchase a buggy from the private sector after the one supplied by the NHS was found not to meet a British Safety Standard. She said:

'In the end we said that we would buy one (buggy), we don't care how much, just get one or we are never going to get out of the door.'

This situation changed in late 1996, when a new NHS voucher scheme was introduced (NHS Executive and DoH, 2000) which aimed to give people more choice by offering 3 options:

1. Present system: to accept the NHS wheelchair prescribed.
2. Independent option: to contribute to the cost of a more expensive wheelchair of the user's choice. The user owns the wheelchair and is responsible for its maintenance and repair.
3. Partnership option: to contribute to the cost of a more expensive wheelchair from a range selected by the local wheelchair service. The NHS owns the wheelchair and is responsible for its maintenance and repair.

In practice, parents said that the voucher scheme was operated differently depending in which area you lived and individual schemes did not always cover powered wheelchairs. This resulted in inequity as some children had been able to purchase a higher specification wheelchair with part-payment by the NHS, whilst others had not.

Under the voucher scheme, some families had also ended up paying unexpectedly high maintenance costs. Mohammed who was a teenager and tetraplegic had opted for the independent option. He had chosen a cutting edge electric wheelchair that he said was the most comfortable and had the longest battery life. The purpose built electric wheelchair cost around £6,000 and was expected to last 5 years. He received a voucher from the NHS for around £2,000 and used his disability living allowance mobility benefit to fund the balance. Mohammed translated for his mum who explained that they could not afford the annual service charge of £600 and each replacement wheel cost £60.70, which was more than a tyre for the family car. Mohammed said that all 4 wheels needed replacing each year and having to pay for the additional costs of maintenance and repair had prevented Mohammed and his family from leasing an adapted van under the Motability scheme. At the time of interview the family could only go out together if Mohammed, his mum and carer travelled in a black cab and his father and siblings travelled in the family car.

The NHS also had strict criteria concerning the supply of powered wheelchairs. For example, Adam was able to use a self-propelled wheelchair, but when he transferred to a large mainstream secondary school he found he was exhausted by lunchtime having propelled himself between morning classes. He was assessed for a powered wheelchair but he did not meet the strict NHS eligibility criteria as he could use, albeit for short distances, a self-propelled chair. His education authority was unable to meet the full cost of a powered chair so like many other children in this qualitative study; Adam turned to Whizz-kidz a non-profit making organisation with charitable status. Whizz-kidz were able to provide Adam with a state of the art powered chair, which he described as *'brilliant'* and *'cool'*.

Many parents expressed disbelief at the cost of providing cycles for their children. Faye was particularly thrilled with her new cycle, which enabled her to join in with her friends on their bikes for the first time. Faye's mum said that Whizz-kidz had provided her 8 year old daughter with a purpose-designed cycle and powered chair. The 2 machines had cost more than the family car (over £11,000) and would cost about the same as the family car to maintain each year.

Whizz-kidz wheelchairs, walkers, buggies and cycles owned by the children in this qualitative study were more expensive than NHS models but had cutting edge design, fast speeds, good manoeuvrability, long battery life, and each machine had been individually tailored and finished with the child's choice of colour and accessories.

New technology

Six children in the current study had a phrenic nerve pacer device implanted which was said by children to confer considerable health gain and improvement in their quality of life (Chapter 4). During exploratory fieldwork, doctors reported that the phrenic nerve pacer was expensive (£29,000) and required a surgical procedure to implant the device followed by 2-3 months as an in-patient learning to use the system. Children still required the same complement of carers and the amount of disposable equipment and supplies increased as children needed supplies both for the phrenic pacer and the ventilator. Parents said that the process of implanting the phrenic pacer was traumatic and they had to cope with the extra costs associated with their child being hospitalised for long periods of time in a specialist unit hundreds of miles from their home.

Another child used a new device to help her cough which her parents had obtained from the United States and purchased themselves at a cost of £3,000. These parents firmly believed that new equipment should be offered to all children and paid for by the NHS.

The extra costs of maintaining the home as a place of employment

All families with employed carers said that there were a lot of extra costs involved when their home was classified as a place of employment. Almost all parents who had employed carers in their home said that they were required to comply with health and safety legislation. This meant that employers had to undertake regular risk assessments and provide such things as fire extinguishers, night-lights and thermos flasks for carrying hot water around the house. Parents also had to pay an increased house insurance premium and declare their home as a place of work.

European Union Working Time Directive

The implementation of the new working time directive had caused a number of problems for NHS managers and parents. Under this directive employees were entitled to regular breaks. In the past, night carers usually worked through the night for 10 hours without any break. Two parents said that they had been informed that in order to comply with the working time directive they would be required to relieve their night carer for a break during the night. One mother said that this would mean having to get up each night from 2-3 am to sit with her child whilst the carer had their statutory break. In her opinion, this arrangement was not appropriate as the carer was employed so that she could get some sleep having cared for her child all day.

Increased household costs

All parents who had employed carers in their homes complained that their household costs had increased dramatically. Parents had purchased such things as fridges, microwaves, crockery and armchairs for carers so that they did not have to intrude into the family kitchen or living area.

Parents said that they had to pay higher electricity bills caused by such things as televisions, lights and other electrical appliances being used at night by carers. Heating bills had also increased as parents were required to keep their houses warm at night for the night carers. Parents also had to budget for increased supplies of toilet rolls, household cleaners, hand soap, milk and biscuits.

Several parents said that they felt obliged to buy their carers presents at Christmas and on the occasion of birthdays. One mother had 6 different carers and she felt this was money she could ill afford; and yet she wanted to be seen as treating her carers well so that they would stay.

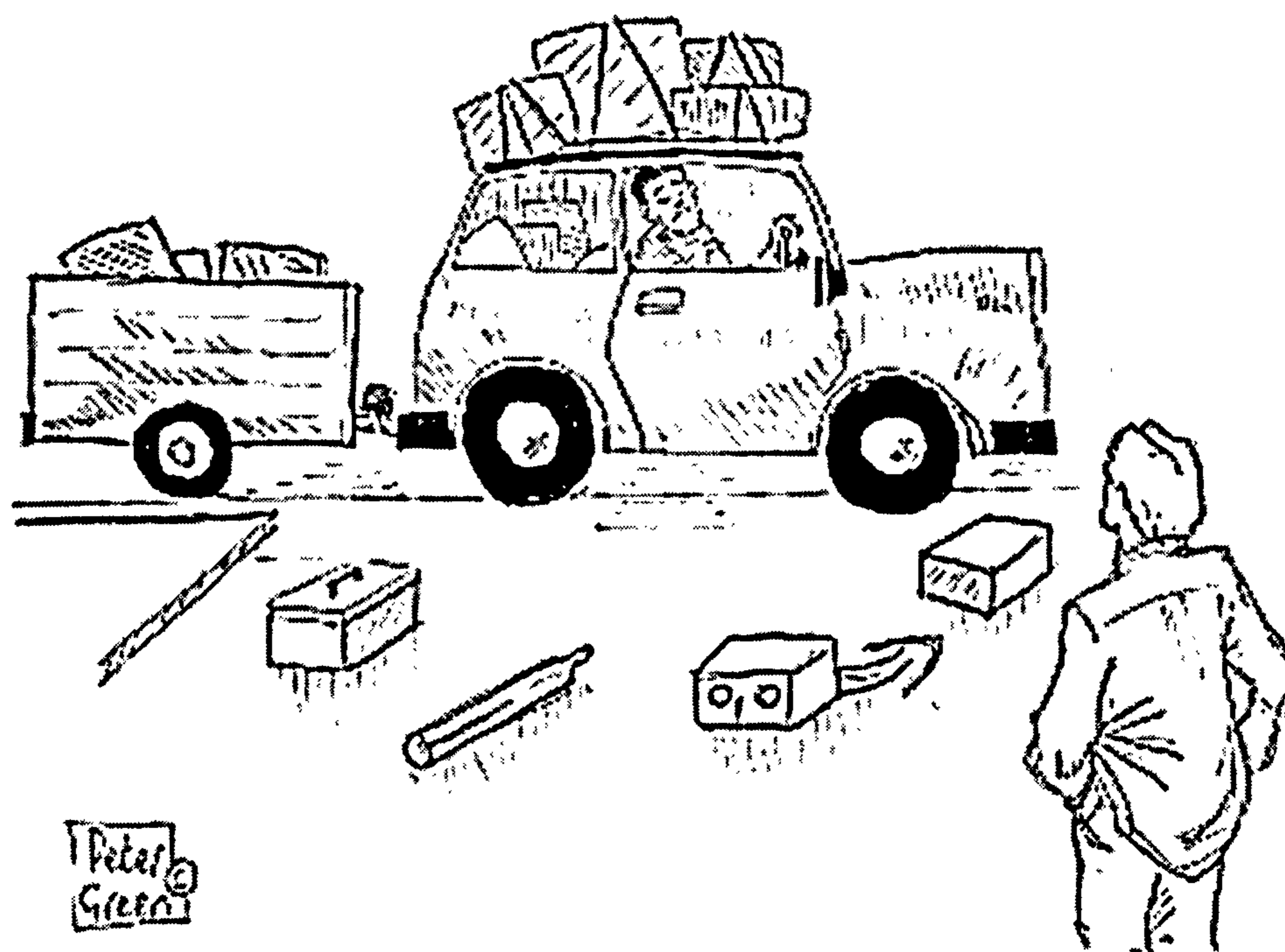
The extra costs associated with getting out and about

Two thirds of children and young people in the sample were unable to use an ordinary car and required specialist transport to get out and about. People carriers and adapted vans were said to be very expensive and parents felt this was a significant financial burden. Not all parents wanted to use the Motability scheme as they would have to return the vehicle if their child died. Some parents preferred to raise money through the media or approach a charity to buy a suitable vehicle. Many families could not go out together until they had acquired suitable transport, which had a negative impact on their quality of life, as highlighted in Chapter 4.

A number of parents mentioned the inflexibility of the mobility component of the disability living allowance. This welfare benefit is paid to help people who have difficulty with getting out and about and who meet strict eligibility criteria. Parents were very unhappy that children age under 3 years of age were not eligible, despite the amount of additional equipment they had to carry around. Parents said they were also turned down if their children could walk. One father had appealed regarding this decision, but had been unsuccessful. This was felt to be unfair as they were not able to get out and about due to the volume of equipment needed.

The cartoonist associated with the study depicted the common experiences described by children regarding their problems with getting out and about with all their equipment (Illustration 7.3).

Illustration 7.3. Common experiences described by children regarding their problems with getting out and about with all their equipment.



In contrast, one teenager was thrilled with his own car that had just been provided through a Motability scheme with an additional grant for specific adaptations from the Government funded specialised vehicles fund. He was currently learning to drive with an additional grant from Motability, which helped to pay for driving lessons, and could not wait to pass his driving test so that he could go wherever he wanted to.

The extra costs associated with starting or returning to school

Three quarters of children who attended school required additional help in the classroom or items of equipment to aid their learning. Although all of the costs were covered by education authorities, some children were very dissatisfied that they could not attend school until assessments of need were made, alterations to the building were completed, classroom assistants were employed and trained, and equipment purchased and installed. Parents said that some schools appeared to look at very expensive building alterations such as installing lifts as opposed to locating the disabled child's class in a room on the ground floor. There was also much dissatisfaction expressed about the

provision of classroom assistants. Almost all children missed some school because classroom assistants were either not in place, were unsuitable, or had frequent sickness or absence from the school.

The extra costs of respite care, breaks and holidays

Respite care

Almost all parents said that being able to take a break away from their caring responsibilities was essential and they felt that their provision for respite care was not sufficient. Not all parents had the provision of respite care built into their care packages and those that did said it was insufficient and inflexible. Most parents with 24 hour care at home said that they also needed time away from their child and the care team. Only one family were able to leave their child and his employed carers in their home whilst they went away on holiday by themselves. In most cases, parents were unable to leave their carers without supervision and house insurance companies would not cover employed carers in the home whilst owners were away.

Breaks and holidays

Almost all ventilator-dependent children, siblings and families who spent their breaks at hospices said that it was fantastic. The children compared hospices with hotels that provided lots of interesting activities and opportunities to mix with other children. Parents liked hospice provision as they felt pampered and could go out socialising knowing that their children were well looked after and having fun.

For almost all parents, taking a family holiday in the UK or abroad became prohibitively expensive if they had to accommodate members of the care team as well. One family had travelled to France with their entire care team of 4 carers. The family had rented an additional villa for the carers and the NHS had paid the carer's travel costs and overtime associated with being away from home for 10 days.

Several families had turned to specialist children's charities who had funded holidays to Disney Land Florida, which was well equipped for managing

disabled children. These specialist charities also arranged and paid for travel insurance, which was otherwise prohibitively expensive for parents.

Nurse managers said that it was also very difficult for employed carers to travel on holiday with families as most had other commitments and responsibilities. Carers also expected to be paid for additional hours spent with families, which would be prohibitively expensive if an entire care team of 6 people required payment 24 hours a day whilst away with the family.

Key theme 3: Social issues concerning 'looked after' children

Three of the young people interviewed were in the care of the local authority, but they were not always aware of the reasons why. A range of complex situations such as child protection and the inability of a parent to care adequately for their children meant that these children and young people were unable to return home.

In these circumstances social workers had to make very difficult decisions about the accommodation of a child where parental care would be competent for a child who is not dependent on a ventilator, but was regarded as insufficiently organised and reliable for a child who is dependent on a ventilator. These are extremely sensitive issues and can bring parents and children into direct conflict with professionals. For example, one mother contested the decisions of doctors and social workers regarding the future accommodation of her child. She eventually fought to have her child discharged to her home with additional nursing support. They have subsequently experienced few problems and the child is thriving alongside his brothers and sisters.

Of the 3 'looked after' children, 2 were living in a residential community and one was in hospital awaiting discharge to a foster placement. A social worker who was interviewed during fieldwork described the considerable costs of managing 'looked after' children. These costs included legal costs, Court time, employing advocates for the child, social worker input as well as the costs of training medical foster carers and providing residential placements.

All the 'looked after' children said that they had spent very long periods of time in hospital. The child who was awaiting discharge to a foster placement said that she had been in hospital for over 4 years and she did not need to be there as she was not ill. In her particular case, no one person was responsible for coordinating her discharge and she was not in contact with her parents who could have advocated on her behalf. The major problem was obtaining sufficient funding to purchase a package of care to enable her to live with her foster parents. Several NHS agencies and local authorities were in dispute over funding and despite the fact that foster parents had been ready for over 3 years, the child remained in hospital. There appeared to be a particular problem in relation to adapting the foster parent's own home; the work had started and then ceased as the firm of builders had not been paid by the local authority. Whilst unresolved funding and housing issues had dragged on there had been 2 attempts to recruit a team of carers to look after the child at the foster parent's home. On each occasion, the team of carers had disbanded as there was no prospect of the child being discharged from hospital as other aspects of the care package were not in place. When the child was eventually discharged from hospital after nearly 6 years, for reasons that were not clear, the foster placement had broken down within months and the child was readmitted to hospital once again.

Key theme 4: The lack of auditing or outcome measures to monitor the effectiveness or appropriateness of the services provided

Although many care packages were reviewed on an annual basis, it was clear from listening to service managers that they were not measured against desirable outcomes for the child and their family, or evaluated regarding the cost-effectiveness or appropriateness of the services provided. Almost all parents said that they felt reviews were undertaken to see if services could be reduced and thus they were a cost saving exercise.

Complaining as a way of getting things done

It became clear from the interviews that almost all parents were dissatisfied with aspects of their care and services and as a result they had made at least one

complaint at some stage in the past. Parents said that not all of their complaints were acted upon. The process of trying to get their child home caused more distress and anger than any other aspect. Parents used a variety of methods to complain and make their views known, including:

- Frequently telephoning people in authority;
- Contacting their Member of Parliament;
- Officially complaining to the NHS Trust or social services department;
- Contacting the Ombudsman, and
- Contacting the newspapers, television and radio.

One parent recalled:

'I had to threaten them. I went to the papers, TV and the radio. It was my MP and the radio... they pushed them (health authority and social services) into doing something.'

Most parents used complaining as a way of getting things done. They would have preferred it if their child's care and services were coordinated more effectively thereby negating the need to complain. Parents found complaining very stressful and the majority of incidents they complained about could have been avoided.

Using legislation and the complaints system to compel the NHS to pay for services has also resulted in a number of unintended consequences; money was said by commissioners and managers to have been diverted from other services to pay for children's home care. During fieldwork, a manager cited the case of a child who had lived in hospital for many years because the money could not be found to provide her with suitable local authority housing and 24 hour nursing care at home. The mother eventually took her case to her Member of Parliament who subsequently put pressure on the health authority and local authority to act. Money was subsequently transferred from services designated for older people to pay for the child's home nursing care and the local authority combined 2 small neighbouring houses into one large dwelling and undertook

extensive adaptations. The manager said that the local authority had spent almost half of its annual budget for housing adaptations on this one child, which meant that other people in the borough would have to wait longer for relatively small but significant adaptations to be completed.

7.3 Discussion

Unnecessary costs caused by prolonged hospitalisation

Ventilator-dependent children and their parents were found to experience additional and sometimes unnecessary financial, emotional and time costs, due to prolonged hospitalisation. These issues have not been explored in any depth in the literature before, and findings provide new evidence on the costs that families endure during months and years of hospitalisation.

Parents and children were firmly of the impression that a package of support services to enable ventilator-dependent children to live at home was cheaper to the NHS than living in hospital and they worried greatly that resources were being wasted by their child's continued hospitalisation.

Parents found there were a lot of additional costs whilst their children lived in hospital. There is a lot written in the literature about extra expenses such as travelling and purchasing meals that parents have to pay for whilst their children are patients (for example, Darbyshire, 1996), but parents in the current study had to endure these costs for months and years at a time. Some parents even had to pay for hospital car park charges and were not always offered residential accommodation to stay with their children. Such charges, or inadequate facilities are in contravention of the Children's Charter which outlines that free residential accommodation should be afforded to at least one parent when their child is in hospital (DoH, 1996).

Discharge management

All parents of children with complex needs for ventilator-dependency expressed their dissatisfaction and frustration at the disorganised and ineffective way that

children were discharged home. Parents suffered considerable economic, emotional and time costs in trying to organise the process when professionals had failed to do so. The barriers to discharge that parents identified are remarkably similar to the studies undertaken in the United States (for example: DeWitt *et al.* 1993, Babinski 1994, Allen *et al.* 1994, Beckett 1996, and Capen and Dedlow, 1998).

Parents as unpaid labour and welfare payments

Families experienced particular financial hardship when they were working as unpaid carers for their children in hospital, which was compounded by lost or reduced income as opportunities for paid employment reduced or ceased. Children in the current studies spent months and years in hospital waiting to be discharged home and the benefits system was not sufficiently flexible to provide sufficient support during this time. Most welfare benefits paid to parents as carers or to parents on behalf of their disabled children did not come on stream until they were discharged home.

The financial impact of living at home with a child with complex needs for health and social care

Findings from the current studies provided a rich description of family life at home and the input of parents as carers and arbiters of care. All of the things identified by the children and their parents as contributing to the costs of care and their dissatisfaction with services are similar to those described in the wider literature for the technology-dependent and disabled generally. In the current studies, ventilator-dependent children on the complex end of the spectrum appeared to need levels of resources that were over and above those of disabled children generally (Roberts and Lawton, 2001). Parents of ventilator-dependent children with particularly complex needs could not manage to care for them without employed carers in the home and a substantial package of support services.

Homes fit for living

Most parents found that their home was unsuitable for their ventilator-dependent child and they needed additional money in the form of mortgages, loans or grants at a time when opportunities for employment were reduced. The time and emotional costs to parents were found to be exacerbated by bureaucratic procedures and means testing of benefits. The financial problems and time costs around adapting or moving house reported here are similar to the contemporaneous findings of Oldman and Beresford (1998 a,b), Bevan (2002) and Beresford and Oldman (2000, 2002).

'Looked after' children.

Few studies have described in detail the experiences of 'looked after' children, especially those living in acute hospitals. Findings make an important contribution to the very small body of evidence on the subject.

Strengths and weaknesses of the qualitative study

Strengths and weaknesses were as described previously in Chapter 4.

Synthesised findings derived from the 4 research themes will be reviewed in an evidence based and policy context in Chapter 9 and further research directions will be outlined in Chapter 10.

7.4 Summary

Chapter 7 continued to focus on the costs of care (theme 3) and satisfaction with services (theme 4) by reviewing the small body of qualitative literature and describing a qualitative study, that aimed to explore the financial impacts to families of caring for ventilator-dependent children, and describe how satisfied parents and children were with the services they received. Findings provide new evidence and add to existing evidence around the substantial financial, emotional, physical, time and social costs to families (especially mothers), and

disorganisation and waste in state-funded services. The Chapter concluded by comparing findings with published literature.

The next chapter will synthesise data around the costs of care (theme 3) and satisfaction with services (theme 4).

Chapter 8

Synthesis of qualitative and quantitative data around costs of care and satisfaction with services

8.0 Introduction

This chapter brings together quantitative findings from Chapter 6 and qualitative findings from Chapter 7, and is the final chapter in the sequence that focuses on reporting and synthesising findings from empirical work around the costs of caring (theme 3), and satisfaction with services (theme 4). A narrative summary technique is used to bring together data around costs and satisfaction from different methodological paradigms to explore and explain similarities and differences around the costs of care and the measurement and experience of satisfaction with services.

8.1 Narrative summary as a method for synthesis

The use of the narrative summary technique as a method of synthesising data was described previously in Chapter 5.

In this chapter the same method of narrative synthesis will be used to juxtapose qualitative findings around the costs of caring and satisfaction with services derived from interviews and or observations with 50 mothers, 17 fathers and 53 children and young people (Chapter 7) against quantitative data derived from an economic evaluation of the costs of care and satisfaction with services with a group of 35 index ventilator-dependent children and their families (Chapter 6).

A narrative summary technique will be used initially to bring together data from different methodological paradigms to explore and explain similarities and

differences in the quantitative and qualitative findings derived from an individual case study. Second, a narrative summary technique will be used to juxtapose qualitative and quantitative findings to explore and explain the outcomes across the entire group of 53 children and young people.

Case study

The similarities and differences between qualitative and quantitative outcomes data concerning the costs of care and perceived satisfaction around receiving complex packages of support services are best illustrated through aligning, in parallel, data derived from a single case study. The qualitative data can then be used to explain and explore quantitative findings in an interpretive way.

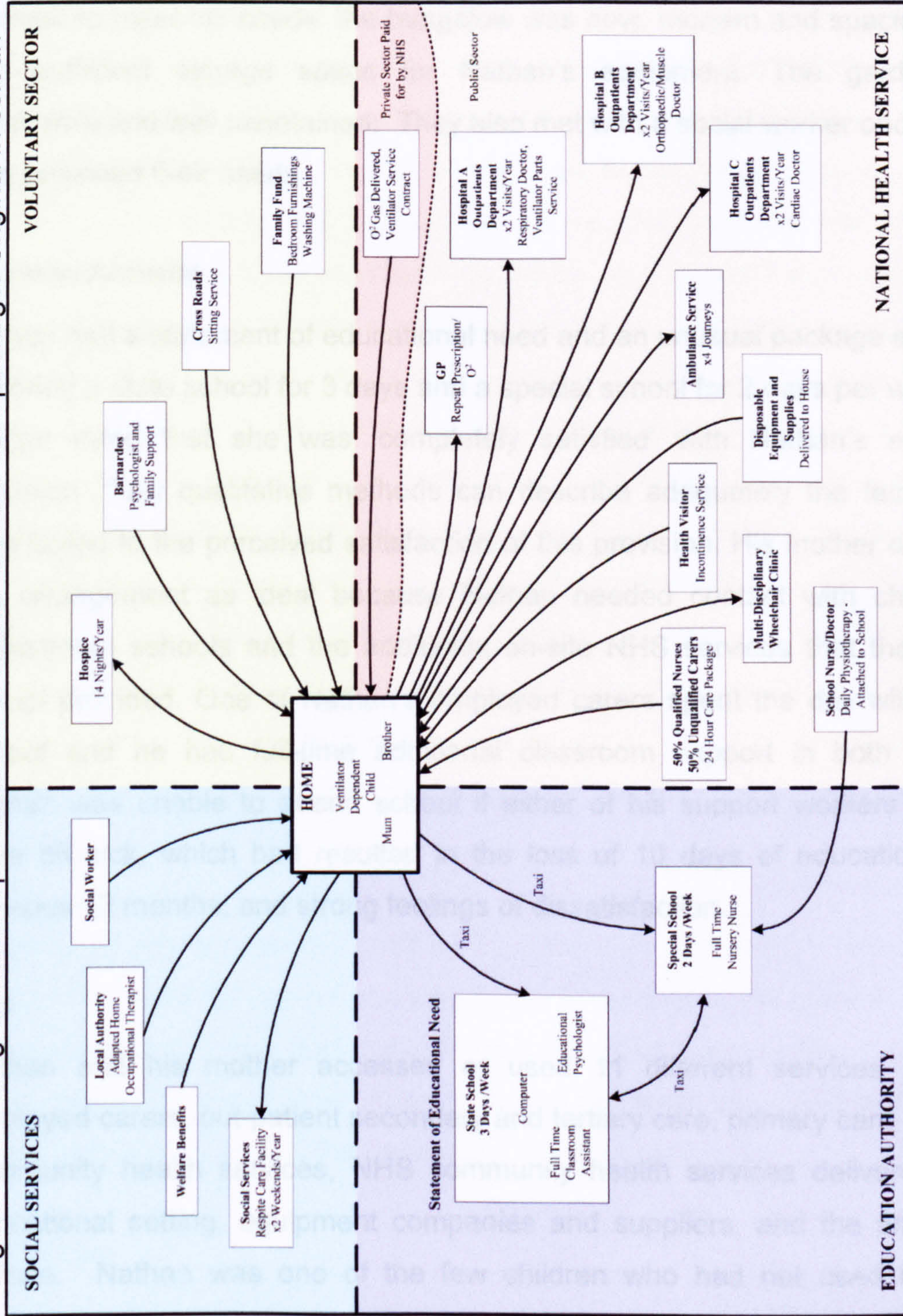
Case Study: Nathan

This case study involves Nathan, a secondary school boy who was dependent on ventilation 24 hours a day following an accident 2 years previously. Nathan was now paralysed from the shoulders downwards and used an electric wheelchair to get around; he needed help with all aspects of personal care. He lived at home with his mum and older brother; his father left the family soon after Nathan's accident.

Nathan's care package

Nathan received a complex and large package of services from the NHS, social services, local education authority and several parts of the voluntary sector. A diagrammatic representation of Nathan's entire care package is shown in Figure 8.1. The total package cost £210,500 for the previous 12 months, with around 80% of costs borne by the NHS (excluding the costs of housing adaptations, domestic transportation and welfare benefits). This is more expensive than the mean cost of packages received by 24 children and young people living at home (mean £104,352 per child per year; range £10,008 to £331,619; SD £84,082).

Figure 8.1. Diagrammatic representation of Nathan's entire care package. Original in colour.



Social Services

Nathan and his family rented a local authority purpose-built bungalow for which his mother received housing benefit to help pay the rent. Nathan rated his accommodation as being 'completely appropriate' on a 5-point Likert scale. Qualitative data provided a rich description as to why the accommodation was deemed to meet his needs: the bungalow was new, modern and spacious, and had sufficient storage space for Nathan's equipment. The garden was accessible and well maintained. They also met with a social worker once a year who reviewed their needs.

Education Authority

Nathan had a statement of educational need and an unusual package in that he attended a state school for 3 days and a special school for 2 days per week. His mother rated that she was 'completely satisfied' with Nathan's education provision. Only qualitative methods can describe adequately the factors that contributed to the perceived satisfaction of this provision. His mother described the arrangement as ideal because Nathan needed contact with children in mainstream schools and the additional on-site NHS services that the special school provided. One of Nathan's employed carers spent the day with him in school and he had full-time additional classroom support in both schools; Nathan was unable to attend school if either of his support workers or carer were off sick, which had resulted in the loss of 10 days of education in the previous 12 months, and strong feelings of dissatisfaction.

NHS

Nathan and his mother accessed or used 11 different services including employed carers, out-patient secondary and tertiary care, primary care services, community health services, NHS community health services delivered in an educational setting, equipment companies and suppliers, and the ambulance service. Nathan was one of the few children who had not used in-patient services during the previous 12 months.

Qualitative interviews were used to explain the resource use that is mapped out in Figure 8.1. Findings revealed several areas of inefficiency that led to feelings

of dissatisfaction. Nathan attended 6 different out-patient appointments at 3 different hospitals each year (on average one appointment every 8 weeks). This meant that he missed 6 days of school unnecessarily and there was much duplication (for example, at every appointment he was weighed and each hospital had a separate set of paper notes).

Nathan had employed carers 24 hours a day (50% qualified; 50% unqualified and a team leader). His employed carers made up the biggest component of the care package (£155,000). Nathan's mum saw herself as a full-time carer and did not mind supervising unqualified carers. Nathan's mother rated the effectiveness of employed care team as 'mixed' on a 5-point Likert scale. Major problems had been encountered with staff sickness, which mainly happened at weekends and night-time, and a high turnover of staff. Carers were limited in what they could do in the home and tasks mostly related to meeting Nathan's needs around his personal care and ventilator. The team leader had attempted to reduce the amount of employed care over the previous 12 months but this had been resisted. Major problems had also been encountered with the delivery of disposable supplies to the home; Nathan regularly ran out of essential supplies. Nathan's mum wanted the carers to be more flexible and fit around her family life. She was dissatisfied with having to chase up the supplies that Nathan needed and said that her telephone bill was eating into her limited budget.

Nathan had one outstanding need for equipment - a new electric wheelchair as he had outgrown his current one which was 3 years old. His current chair was uncomfortable and had poor battery life and he had been waiting 6 months to be measured for a new chair. Apart from that his health was good and all of his health needs were met by the package of services provided.

Respite care

Despite having a package of 24 hour employed care and plenty of space at home, Nathan's mum identified a need for more in and out of home respite care so that she could have a break from being around carers. She had herself

approached her social worker and 3 charities to arrange respite care; this provision was still said to be inadequate.

Family income

The family received an income of between £10,000 to £15,000 per year. This was made up entirely of benefits and no child maintenance payments had been received from Nathan's father. This placed them well below the national average wage in 2002. The Family Fund had provided money for extra bedroom furnishings and a washing machine. Nathan's mum said that money was tight but they had a comfortable home with all the fixtures and fittings they wanted (television, hi-fi, computer games, etc). Her budget did not stretch to spending much time away from the family home and she was dissatisfied at having to pay additional heating costs as the employed carers got cold at night.

Social needs

Nathan rated his social life as 'mixed' on a 5-point Likert scale. He said that he spent too much time in the company of adults; he did not get out enough and he wanted his carers to take him to meet more friends and participate in after school clubs and sports. He wanted to go to Disney Land in Florida and attend football matches regularly. Nathan's mum said that she wanted more time to herself and to be recognised as a person in her own right. Nathan's brother felt very left out and could exhibit challenging behaviour. The care team had to adhere to strict risk management policies, which limited the activities they could participate in, and as a single parent family they did not have the resources to undertake activities without the support of carers.

Nathan rated the transportation available to him as 'mixed' on a 5-point Likert scale. He said he was dissatisfied with the transport options available to his family as Nathan's mother did not drive and could not afford driving lessons. Nathan used the mobility component of the disability living allowance to pay for expensive black cabs when he wanted to go out; this made spontaneity difficult and his allowance was quickly used. His mum said that they also liked to walk to local places of interest (Nathan propelled himself in his electric wheelchair,

although he could not go far because his chair was not comfortable and had a poor battery life). Nathan said that if the family had the money to buy an adapted people carrier then they could go out to more places as a family, but only if the employed carer was cleared by her manager in terms of risk management.

Coordination of the entire care package

Nathan and his mum rated the coordination of their care package as being 'completely dissatisfied'. Parallel qualitative data revealed that too many organisations were involved and too many people visited their home. Nathan's mum was the only person with a complete picture of the entire care package and it was her responsibility to communicate between all the component parts; things did not always run smoothly and she found this very stressful. When she voiced concerns that her needs were not being met, the response was to provide more services rather than increase the effectiveness of current services. As a result her doctor had recently prescribed anti-depressants as she found it difficult to cope. Nathan's mum wanted more in and out of home respite so that she could get away from the stress of having to deal with so many people.

In synthesising findings across studies, it was possible to determine that this package of care (which cost £210,500 and was one of the more expensive packages) had aspects that were high quality and generated feelings of satisfaction, aspects that were low quality and not focused around the child and generated feelings of dissatisfaction, and areas of gross inefficiency which caused a significant amount of distress to the very people that the services were designed to support. In synthesising quantitative and qualitative findings it is easy to see that for Nathan and his mother the quality of care and services received were synonymous with their own quality of life, and dissatisfaction with the coordination and effectiveness of services received was an important component of their life dissatisfaction and inability to cope.

There are a number of changes that could be made to improve the cost-effectiveness and appropriateness of the package of support services and bring

about the desired outcomes for Nathan and his family. Changes likely to bring about an improvement to the social benefits of packages of support services will be discussed further in Chapter 9.

Synthesis of qualitative and quantitative data around costs of care and satisfaction with services across the entire group of children

A narrative synthesis of qualitative findings around costs and satisfaction with services derived from interviews and or observations with 50 mothers, 17 fathers and 53 children and young people (Chapter 7) will be juxtaposed against the quantitative costing and satisfaction data derived from the entire group of 35 case studies of index children (Chapter 6). Synthesis of findings across studies will firstly focus on children living in hospital and then children living at home.

Narrative synthesis of qualitative and quantitative data around 7 children and young people living in hospital for the previous 12 months.

The total cost of the support received by these 7 ventilator-dependent children and young people was over £3.3 million, with 98% of costs borne by the NHS. The average cost of services used by each child was £482,259 for the previous 12 months (range £335,489 to £633,732; SD £100,576). All parents and/or children scored that they were either very or completely dissatisfied, or had mixed feelings about the appropriateness of living in hospital.

The children either lived in intensive care or high dependency environments; the cost of in-patient care consumed 92% of the total resource use and was the most expensive placement. In contrast, parallel qualitative findings revealed most of the children and young people lived in hospital long after their medical needs had been attended to and none of them wanted to be there. Children also experienced several moves between hospitals, some of which were located hundreds of miles from the family home which increased travelling and time costs for parents. Parents had to cope with many unexpected and some unacceptable costs and charges whilst their children lived in hospitals. These costs ranged from loss of earnings to additional travel, accommodation, food and child minding costs. Hospitals were not always meeting their obligations in

terms of providing free residential accommodation to parents to stay with their children. Welfare payments to reimburse travel costs were only paid to those on low incomes and most parents with modest incomes were above the payment threshold. Children were not eligible for welfare benefits such as the disability living allowance until after discharge home, which meant that around the time of greatest need families could face financial hardship, as entitlements to employers' sick pay diminished or parents had to reduce or give up work.

All of the children required significant input from social workers and nurses in order to organise their discharge home; the costs to social services were £37,945 and nursing costs were consumed by the cost of the bed per day (mean £1,700 per child). Parallel qualitative findings revealed that children had to wait months and in some cases years to be discharged home because of financial and organisational inefficiencies that became insurmountable barriers to discharge.

Two of the children had their own employed care teams learning to look after them in hospital before being discharged home, which came to £106,481. This meant that the period of time prior to the child's discharge from hospital was the most expensive, as both ward nurses and the child's own carers were being employed at the same time. Parallel qualitative findings found that although nurses and social workers spent a lot of time organising meetings, they lacked sufficient authority to work across agencies and sectors, and neither had the time to take sole responsibility for organising the discharge process or institute effective recruitment practices. Lack of a key person to coordinate the discharge process and lack of multi-agency working were found to act as major barriers to discharge. As a result, gross inefficiencies were identified around the time of discharge, which prolonged the process and coincided with a period of additional resource use, if employed carers were being recruited and trained ready to care for the child once discharged home.

The other costs incurred whilst children lived in hospital settings were due to the educational needs of the children; one child was attending a mainstream primary school and required a taxi to school each day and additional support in

the classroom; the costs of extra educational services received were £26,809. Parallel qualitative findings indicated that many children living in hospitals missed out on chunks of their education as provision was insufficient to meet their needs.

Narrative synthesis of qualitative and quantitative data around 24 children and young people living home for the previous 12 months.

The total cost of the support received by 24 ventilator-dependent children and young people was over £2.5 million (excluding the cost of adapting housing for a disabled child and the cost of domestic transportation) with 83% of costs borne by the NHS. The average costs of services used by ventilator-dependent children and young people were £104,352 per year (range £10,008 to £331,619; SD £84,082).

'Simple' versus 'complex' ventilator-dependency

Quantitative findings revealed a group of children who had less complex needs around ventilator-dependency. For this group of children with 'simple' ventilator-dependency and moderate associated impairments, the average total support costs per child per year were £17,876. In contrast, for the remaining 19 children and young people with more 'complex' needs, the average total support costs per child per year was £127,109, over 7 times greater.

Parallel qualitative findings were able to confirm that children and young people with simple needs around ventilator-dependency were able largely to manage their care without additional services, such as employed care in the home, and they were satisfied with their treatment. In contrast, children with more complex needs around ventilator-dependency had the most need for support services and expressed the higher levels of dissatisfaction (as illustrated in the case study concerning Nathan).

Multi-agency working and management of care packages

Ventilator-dependent children used considerably more health-related services and resources than normative population groups and young disabled people

with hemiplegic cerebral palsy. Only 45% of respondents were satisfied with the coordination of services. Hospital services absorbed 17.8% of the total annual budget (£444,994) with 97% being spent on in-patient care and the remainder on a range of out-patient and therapy appointments. Six children had a phrenic nerve pacing device implanted at a cost of around £36,000 per child; the long-run marginal cost of phrenic nerve pacing was £6,670 per year plus £500 for disposable supplies per year.

Parallel qualitative data revealed that hospital admissions were mainly due to acute illness or for major surgery and the implantation of new expensive technology, which did not reduce their overall resource use. Parents had to cope with the additional costs associated with their child's hospitalisations for prolonged periods of time (travel and child minding).

Parents and children complained about the multitude of appointments that children attended. Most hospital-based out-patient services were not organised specifically for children and young people dependent on ventilators, so families were sent to a myriad of individual doctors and therapists in different locations as opposed to the child and their family visiting a multi-disciplinary team in one clinic on one day. This lack of coordination resulted in duplication of NHS resources and additional travel costs for parents and lost employment.

Families were also in contact with a number of different services in the community and reported difficulties in negotiating their way into and around services and understanding the roles and responsibilities of professionals. Many of the children with complex ventilator-dependency received relatively small interventions from a multitude of professionals; this resulted in family homes being disrupted constantly by people calling at the house to deliver each individual facet of therapy or care. This is both wasteful in terms of staff travelling time and unsatisfactory to children and their families in terms of time costs.

Employed care teams

Children with the most expensive support packages had employed care teams with a high ratio of trained nurses (mean cost £57,407 per child per year; range £0 - £181,126; SD £63,710); 84% had experienced problems with their employed care teams.

Parallel qualitative data revealed that professionals tended to encourage parents to have unqualified carers in their homes as they were cheaper than qualified nurses. The consequence of this arrangement was that there was an expectation that a family member (usually mothers) would either constantly supervise and/or provide back-up to the carer; these roles had considerable time consequences for some mothers who also experienced reduced employment opportunities outside of the home. Some families preferred having unqualified carers in their home as they were more flexible than qualified nurses and tended to stay longer, which reduced recruitment and training costs. The major problems encountered were staff sickness and turnover, and inefficient management practices.

Equipment

All children required medical devices and equipment costing on average £7,220 per child per year (range £1,894 - £20,186; SD £4,689).

NHS Trusts did not always procure equipment in the most cost-effective manner. The provision of wheelchairs was singled out as being inefficient and the voucher scheme introduced inequity especially for low-income families.

Inappropriate use of supplies

Children consumed varying amounts of disposable equipment and supplies (range £1,000 – £15,876) with an average cost of £8,133 per child, which absorbed 8% of the total budget; 60% of parents reported significant problems with obtaining supplies.

Parallel qualitative findings confirmed the nature of the problems with the supply and delivery of items to domestic homes. Parents wasted a lot of time ordering supplies from various agencies and chasing up lost deliveries. Most parents were very careful concerning the judicious use of disposable supplies, but a minority of parents used excessive amounts of some products due to lack of information or expertise.

Housing

Accurate figures were not available regarding the cost to the state of providing local authority accommodation or means tested disabled facilities grants.

Parallel qualitative findings revealed that parents experienced considerable cost and time burdens around modifying or moving house. The process of applying for a grant was complex and unfair. Parents felt that their income should not be taken into account.

School

Eighteen ventilator-dependent children and young people required additional support in the classroom and or transport to school; this absorbed 7% of the total budget (Mean £7,143 per child per year); 59% of respondents were satisfied with educational services.

Assessments of need were slow and children could not attend school until resources were provided. Schools did not always consider cost-effective alternatives. Attendance at school was affected by ineffective services; especially concerning transport, sickness or absence or classroom support.

Costs of hospital versus home care

It was mostly but not always cheaper for a ventilator-dependent child to live at home; a care package including qualified nurses and 24 hour care (£239,855) was more expensive than a children's ward (£155,158) and residential placement (£105,555 to £194,351) but less expensive than a long-term ventilation unit (£301,888) and intensive care unit (£630,388) where most

ventilator-dependent children live whilst in hospital. The cheapest option was care by parent(s) at home (£46,483 per year).

When quantitative cost estimates were juxtaposed alongside qualitative findings, it was found that parents, children, and most doctors perceived that living in a hospital bed was the most expensive option compared with providing a package of support services to enable the child to live at home. Only the commissioners of services and NHS managers had an understanding of the wider economic situation concerning block contracts to provide intensive care services and the additional costs required to commission new services to enable ventilator-dependent children to live at home.

8.2 Discussion

The qualitative and quantitative data synthesised in this chapter were derived from studies embedded in different philosophical paradigms and underpinned by different theoretical perspectives. The quantitative estimation of costs was underpinned by the medical model of disability and measured the resource use and costs of care for ventilator-dependent children against costs incurred by 'normal' children in the population; differences in resource use were interpreted as an indication of increased requirement for support from the state. In contrast, the qualitative exploration of the costs of caring for children dependent on ventilators was underpinned by the social model of disability and provided rich data around the financial, physical, emotional and time costs of care to parents. The qualitative data are particularly important in this respect as it was not possible to estimate the costs of parental input in the quantitative economic evaluation.

Cost-effectiveness of service delivery and organisation

Evidence from current studies identified that a large financial (albeit insufficient) state-funded investment had been made in purchasing services and resources for ventilator-dependent children. The potential cost-effectiveness of services was not realised due to factors such as poor organisation and delivery. Examples of excessive waste were identified around ineffective multi-agency

working, particularly around discharge management, and the delivery and supply of disposable equipment to domestic homes. Most families had not received information about independent brokerage schemes.

Multi-agency working

There is a well-established body of literature that addresses the question of what makes a successful and cost-effective partnership in terms of delivering multi-agency services. According to a recent study by Townsley *et al.* (2004), research is no longer coming up with any new evidence concerning the factors that lead to successful cost-effective partnerships between organisations engaged in delivering packages of support services. Townsley *et al.* (2004) summarised the common success factors to effective multi-agency working, as follows:

- Explicit agreement about how the partnership will pool or share resources such as time, people and money;
- Explicit agreement to a clear, shared vision that defines the purpose of the partnership, and common objectives for achieving that vision;
- A clearly defined structure or model to explain how the multi-agency process will operate, particularly in terms of the nature of the work with children and their families and the expected outcomes for them;
- Clarification of the roles, responsibilities and contributions of the people involved in the multi-agency process;
- Effective leadership and the existence of allies and champions at strategic and operational levels within all organisations involved;
- The provision of opportunities for learning, support and supervision;
- Clear and agreed management and accountability;
- Good communication between all those involved;
- Partnership with children and families;
- Regular monitoring and evaluation, and
- Clarity about timescales and future planning.

Evidence from the current studies indicates that for around half of the children and young people with the most complex needs, many of these success factors were not evident in the management and delivery of their resources and services.

Synthesis of findings across studies revealed that the period around discharge from hospital was the most costly in terms of resource use and the most inefficient in terms of service organisation and delivery for both the state and parents. Having a single key identified person to manage discharge from hospital and a similar person to manage the package of support services in the community was found to be beneficial in terms of maximising the effectiveness of the care package (Muckherjee *et al.* 1999; Greco *et al.* 2004).

Financial welfare of families

It was not possible to estimate the costs of parental input but qualitative findings helped to fill this gap by providing a detailed description of the input of parents and their additional expenditure. Findings add to existing qualitative evidence that the savings to the state of home care could be at the expense of parents who endured additional economic and time consequences, especially around reduced opportunities for employment. Synthesis of findings across studies confirm that the resource use consumed by ventilator-dependent children is far higher than that described in the general literature around disability and the financial impacts to parents are particularly high when their children live in hospital and when employed carers work in the home.

Synthesised findings derived from the 4 research themes will be presented in the next chapter and further research directions will be outlined in Chapter 10.

8.3 Summary

This chapter brought together quantitative and qualitative findings around the costs of caring (theme 3), and satisfaction with services (theme 4), derived from Chapters 6 and 7, by using a narrative summary technique.

Synthesis of findings across studies enumerated the large (albeit insufficient) state-funded financial investment in services and resources for ventilator-dependent children and linked parallel qualitative data concerning specific aspects of poor organisation and ineffective resource use (particularly around the discharge management of children from hospital, and the lack of multi-agency working across sectors in the community). Explicit linkages were made between the skill-mix of employed care teams and the negative time and economic impacts on families (especially on mothers). The chapter concluded by reviewing the success factors for multi-agency working and issues around the financial welfare of families who care for ventilator-dependent children.

The next chapter draws together empirical data around the 4 research themes reported in Chapters 2-8 in order to compare the costs of caring for ventilator-dependent children with the social benefits within an evidence based and policy context.

Chapter 9

Synthesising the results within an evidence based and policy context

9.0 Introduction

This chapter has three distinct sections.

First, a synthesis of findings concerning the costs of caring for ventilator-dependent children in different settings are compared with the social benefits. In order to structure the synthesis, findings are assessed against a set of criteria for framing evidence around health problems and medical conditions contained in the new National Service Framework for Children (DoH, 2003d).

Second, one of the major conceptual issues to emerge from the synthesis was the lack of clarity regarding the health and social needs of children and their families, and the outcomes of the care and services provided. This issue is discussed in further detail as it has important practical implications for the commissioning of services based on needs, costs and outcomes.

Finally, synthesised findings are reviewed in the light of current health and social policy initiatives.

9.1 Synthesis of findings concerning the costs and social benefits of caring for ventilator-dependent children

The aim of the research programme reported in this thesis was to provide rigorous evidence where it has not existed before, concerning the costs and social benefits of caring for children dependent on ventilators. In this section,

results from the different studies under the 4 research themes are brought together. The 4 research themes are as follows:

1. The origins and size of the population;
2. The health-related quality of life of children;
3. The costs of caring, and
4. Satisfaction with services.

Synthesised findings will be assessed against a recently published set of criteria contained within the National Service Framework for Children in Wales (DoH, 2003d). The purpose of these criteria is to outline the evidence base required to underpin the development and funding of appropriate multi-disciplinary services, based on needs, costs and outcomes. These criteria are summarised in Figure 9.3, and the main headings will be used to structure the synthesis of findings across the 4 research themes.

Summary of research conducted under the 4 research themes

The origins and size of the population (theme1)

Two studies were undertaken (one qualitative, one quantitative) to determine the origin and enumerate the size of the population of children and young people dependent on ventilators in the UK (theme 1). A summary of the research designs employed and research questions addressed is outlined in Figure 9.1.

Figure 9.1. The origins and the size of the population (theme 1).

Research Methods	Research Questions
Quantitative Secondary analysis of NHS Hospital Episode Statistics and Scottish Morbidity Records of paediatric admissions to hospital.	Can the population of children and young people who are dependent on ventilators be enumerated from routinely collected NHS data, and if so, how many children are there and what resources do they consume?
Qualitative Exploratory observational fieldwork including qualitative interviews with doctors, nurses, rehabilitation professionals, service commissioners and managers.	How and why has this population emerged?

The costs and social benefits of long-term ventilation in children (themes, 2,3 and 4)

Four studies were undertaken (2 qualitative, 2 quantitative) to determine the costs and social benefits of long-term ventilation as an intervention for children and young people in the UK (themes 2, 3 and 4). The research designs employed and research questions addressed are summarised in Figure 9.2.

Figure 9.2. Summary of studies concerning themes 2,3 and 4.

The costs of caring for children and young people dependent on ventilators (Theme 3)	
Research Method	Research Questions
<p>Quantitative Economic evaluation of the support costs and resource use of 35 ventilator-dependent children over the previous 12 months</p>	<p>What are the long-run marginal service costs of supporting ventilator-dependent children in hospital, home and residential settings over 12 months?</p>
<p>Qualitative Qualitative interviews with 70 parents and 53 ventilator-dependent children and young people.</p> <p>Exploratory observational fieldwork involving doctors, nurses, rehabilitation professionals, service commissioners and managers.</p>	<p>What are the financial impacts of caring for ventilator-dependent children?</p>
The social benefits of long-term ventilation in children: The health-related quality of life of children and young people dependent on ventilators (Theme 2)	
<p>Quantitative Measurement of health-related quality of life with EQ-5D and KINDL with 28 parents and 17 ventilator-dependent children. Findings were compared with normative data.</p>	<ol style="list-style-type: none"> 1. Do ventilator-dependent children have lower health-related quality of life than a normative population of school children? 2. Do ventilator-dependent children's self-reports of their health-related quality of life differ from those of their parents? 3. Does the health-related quality of life of ventilator-dependent children living in hospital differ from ventilator-dependent children living at home?
<p>Qualitative Qualitative interviews with 70 parents and 53 ventilator-dependent children and young people.</p> <p>Exploratory observational fieldwork involving doctors, nurses, rehabilitation professionals, service commissioners and managers.</p>	<p>How do ventilator-dependent children and their parent(s) view the child's health and quality of life and what impacts do health, social and education services have on their quality of life?</p>
The social benefits of long-term ventilation in children: Satisfaction with services and unmet needs (Theme 4)	
<p>Quantitative Satisfaction with services was measured with questions requiring answers on a 5 point Likert-scale delivered to 35 ventilator-dependent children and or their parent(s). Unmet needs identified on a nominal scale and brief description provided.</p>	<p>How satisfied are parents and children with the services they receive?</p>
<p>Qualitative Qualitative interviews with 70 parent(s) and 53 ventilator-dependent children and young people.</p> <p>Exploratory observational fieldwork involving doctors, nurses, rehabilitation professionals, service commissioners and managers.</p>	<p>How satisfied are parents and children with the services they receive?</p>

Structuring the synthesised findings against the National Service Framework for Children criteria

The criteria proposed in the National Service Framework for Children provide a framework for evidence concerning specific health conditions (DoH, 2003d). The framework is designed to assemble evidence from the point of need through to implementation of identified effective interventions. It is devised for general child services and provides a useful background in which the results from the studies undertaken for the thesis can be placed and assessed. The synthesised findings will be matched against the following criteria outlined in Figure 9.3:

- Demonstrable need and predicted increase in need;
- Scope for health benefit, including magnitude of benefit and effectiveness and cost-effectiveness of interventions;
- Effectiveness of service models for implementing intervention(s);
- Areas where existing services are not meeting needs or where there is variation in the quality of services provided;
- Practicality of implementation, including organisational capacity, workforce implications, and acceptability to children, young people and their families, and
- Measurability of implementation, including practicality of setting up new monitoring arrangements.

Figure 9.3. Criteria for framing evidence around health problems and medical conditions set out in the new National Service Framework for Children (DoH, 2003d).

Demonstrable need, for example:

- Numbers of children and/or young people affected
- Impact on affected individuals
- Cost to individual, the NHS, society

Predicted increase in need, for example:

- Increasing prevalence/incidence of the condition/problem
- Increasing prevalence of the risk factors for developing the condition/problem

Areas where existing services are not meeting needs or where there is variation in the quality of services provided, in terms of, for example:

- Access to services
- User and carer experience
- Quality of care/service provided
- Patient outcomes

Scope for health benefit

- Effective intervention(s) available
- Effectiveness of intervention(s)
- Cost effectiveness of intervention(s)
- Population subgroups likely to benefit (NB different needs of infants, children and young people; looked after children; children with a physical, learning or sensory disability; children and young people living in residential care and other institutional settings; young people living in custodial settings; children and young people from different ethnic groups; children and young people who are socially deprived; etc)
- Number/proportion of children and or/young people likely to benefit
- Magnitude of benefit

Effectiveness of service models for implementing intervention(s):

- Effectiveness of service models for implementing intervention(s)
- Cost-effectiveness of alternative service models

Practicality of implementation in terms of:

- Organisational capacity
- Cost of implementation
- Workforce implications
- Integration with other initiatives – and potential for synergy
- Acceptability to children and young people, and their families
- Acceptability to health professionals
- Levers for change

Measurability of implementation:

- Robustness of existing monitoring arrangements, including available indicators for both process and outcome
- Practicality of setting up new monitoring arrangements

Demonstrable need and predicted increase in need

The findings from the theme concerning the origins and size of the population help to address some of the criteria concerning the demonstrable need and whether it is increasing.

Findings revealed that using routinely collected NHS data, it was not possible to enumerate the numbers of ventilator-dependent children in the UK. Thus far, it has not been possible to estimate incidence and prevalence with sufficient methodological rigour. Although known to be a gross underestimate, the only available national figures to date come from a postal survey of clinicians undertaken in 1996-7, which identified 93 children living at home and 43 children living in hospitals (Jardine *et al.* 1999).

Qualitative research with doctors, commissioners of services and nurses found that increasing numbers of ventilator-dependent children were being managed each year, thereby confirming the trend seen in the point prevalence surveys. The emergence of this population was said to be due to 5 factors, including:

1. New improved portable technology;
2. Improved intensive care and increased survival of children;
3. Legal guidance on withholding and withdrawal of treatment;
4. The impact of parents and advocacy groups campaigning for children to be treated, and
5. The emergence of obesity (a new lifestyle disease).

This research also however highlights the difficulty of evaluating a group of children not defined by specific diagnostic codes, but by dependency on a common piece of medical equipment – the ventilator.

Scope for health benefit

The findings from the 3 research themes concerning the costs and benefits of long-term ventilation provide evidence to address some of the criteria including the magnitude of benefit, and the effectiveness and cost effectiveness of interventions. Evidence around population subgroups is included under the criteria concerning areas of existing services that are not meeting needs.

Magnitude of benefit

Long-term ventilation is currently used as an intervention to treat children and young people with a range of diagnoses and clinical states; ranging from children in persistent vegetative states who live in hospital wards, through to young people who are about to enter the workforce, having completed their higher education. Economic constraints concerning the allocation of finite budgets have driven the need to define health-related quality of life in ventilator-dependent children, in order to generate evidence of effectiveness and delineate the magnitude of benefit.

Long-term ventilation either improved or maintained life and as such conferred a benefit to children's health; although in the case of children with degenerative conditions the benefit was confined to the short to medium term as a palliative intervention towards the end of life. The magnitude of benefit could not be determined with current research methods for all groups of children, especially those with profound sensory impairments.

Ventilator-dependent children and young people had different perspectives regarding their health and quality of life than their parents; children on the whole perceived their health and quality of life to be better. There was consensus between parents and children that quality of life equated to feeling well, and being able to enjoy an 'ordinary' childhood with lots of varied experiences. Ventilator-dependent children as a group had lower self-esteem than children in the general population. Many of the things that affected their self-esteem were beyond their control, such as how society viewed disability in a negative way; the sometimes unnecessary constraints and lack of control children had over

their lives, and the negative impact of inadequate and sometimes poor quality services.

Effectiveness and cost-effectiveness of interventions

The costs of long-term ventilation arose from decisions to commit (or not commit) resources to the treatment of underlying disease and to interventions that minimise the disabling barriers that resulted from children's impairments. The health and social benefits of long-term ventilation were only fully realised if children and young people were provided with resources to live at home with their families and the disabling barriers to social exclusion were removed.

The costs of caring for children dependent on ventilators were high to taxpayers (mean £179,408 per child per year; range £10,008 - £633,732); 92% of annual support costs were funded by the NHS. The financial and time costs were high to parents although figures were not calculated. The location of care had significant impacts on both costs and social benefits. These will be described in relation to each placement.

Hospital placements varied in cost per child per year from intensive care unit (£630,388), high dependency unit (£301,888) to children's ward (£155,158) but conferred the least social benefits to children and their families (especially intensive care), and significant additional financial and time costs to parents.

The cost of supporting children in their own homes per year ranged from £239,855 (24 hour care with qualified nurses) to £46,483 (parents as unpaid carers). Living at home conferred significant social benefits to children and their families, if services were of good quality, effective and child-centred. Parents faced increased costs around caring for a disabled child and when their home became a place of employment; mothers in particular experienced reduced opportunities for employment as caring responsibilities increased or if they were required to supervise unqualified carers. The time consequences of care on mothers conferred many social disbenefits and feelings of dissatisfaction.

Children with complex needs around ventilator-dependency consumed 7 times more resources at home than children with simple needs (mean £127,109 versus £17,876 per child per year). The latter group with simple needs around ventilator-dependency had fewer requirements for services and most experienced considerable health gain and social benefit from long-term ventilation. These children and their families were far more satisfied with their quality of life and inclusion in society in general.

Six children and young people in the current studies had benefited from the implantation of a phrenic nerve pacing device, and 2 were awaiting surgery to implant the device. This new and expensive adjunctive technology added to the costs of care and resources consumed by children as the phrenic nerve pacer was used in addition to their existing ventilators and they required the same number of carers. In addition to the costs of implanting the device (around £36,000), the long-run marginal cost of phrenic nerve pacing was £6,670 per child per year plus £500 for disposable supplies. Parents had to absorb additional costs and loss of employment opportunities due to a prolonged period of hospitalisation (2-3 months) when the technology was implanted.

This new technology, however, conferred considerable social benefits from the perspective of the child including; being able to spend time off the ventilator during the day; improved cosmesis; a louder voice; more opportunities for socialising with other children; and, reduced stigma due to the equipment being less visible and emitting less noise.

Effectiveness of service models for implementing interventions

Findings from the 3 themes concerning the costs and benefits of long-term ventilation provide evidence to address the criteria concerning the effectiveness and cost-effectiveness of service models.

Cost wise, it was mostly, but not always cheaper to provide a package of support services for a child to live at home (mean £104,352 per child per year (range £10,008 to £331,619; SD £84,082). But, contractual arrangements around the provision of intensive care acted as a perverse financial incentive to

keeping these children in hospital, because providing a package of community support increased immediate financial demands that did not exist previously. Children who were kept in hospital unnecessarily experienced the worst social outcomes, as did their families.

Areas where existing services are not meeting need or where there is variation in the quality of services provided

Findings from the 3 research themes relating to the costs and benefits of long-term ventilation provide evidence that address the criteria concerning access to services, user and care experience, quality of care/services provided and patient outcomes. As all of these issues are associated with poor quality services, findings will be presented according to the services identified as not meeting needs.

Hospital discharge management

The social benefits of care were maximised if children were discharged from hospital within a reasonable length of time. Children with the most complex needs however spent too long in hospital waiting to be discharged home. Costs to the NHS could be very high at this time if children also had employed care teams in place. Additional human resources were not always made available to ensure that the discharge process was efficient and cost-effective. The financial barriers to providing packages of services that enable children to be discharged home are only likely to be resolved with greater political and managerial intervention. The inability of the NHS and social services to bring about the discharge of children resulted in significant psychological and financial disbenefits for children and families (especially in relation to eligibility to welfare benefits).

Despite a substantial evidence base, and the availability of published clinical guidance around discharging ventilator-dependent children from hospital (for example, Spence, 1995; Jardine and Wallace, 1998; NHS Executive North West, 2000), findings suggest that such guidance is not translating into effective practice. Almost all NHS Trusts were found to be ill equipped in organising the

discharge of ventilator-dependent children from hospital. Children who are dependent on ventilators constitute a low volume, yet high cost heterogeneous group and it is not surprising that nurses have little experience of organising such a complex discharge process. There is evidence that organising the discharge of a patient with complex needs constitutes a huge and hidden workload for nurses (McWilliam and Wong, 1994). There is an urgent need for a consistent national approach to address the shortcomings of the present system of discharging these children (McGinley *et al.* 1996); evidence in the literature suggests that the employment of discharge coordinators increases the efficiency of the discharge process (Raulin and Shannon, 1986; Yaksic *et al.* 1996; Piskule and Johnson, 1997; DoH, 1999a).

Organisation and management of community support services

There was a high cost to the state of providing support services to enable children to live at home (mean £104,352 per child per year (range £10,008 to £331,619; SD £84,082). Good quality, flexible services delivered by multi-agency partnerships and coordinated by a key worker were associated with increased social benefits, such as positive experiences of wellbeing and positive feelings of satisfaction, for both children and their parents. Additional benefits were derived from access to high quality services that addressed the health and social needs of the child and their entire family.

Despite the high cost to the taxpayer, only around half of packages of support services brought about the desired social benefits as identified by the child and their family. In particular, children did not gain maximum social benefit from the investment in resources if their communication needs had not been addressed as they were unable to articulate their needs or engage socially with others. Parents considered that the family's social needs were equally important as the health needs of the child and believed that services provided needed to address both in order to be effective. Faced with limited budgets and demand that constantly outstripped supply, NHS managers tended to concentrate on meeting the health-related needs of children for which their services were primarily commissioned to address. Services were cut if reviews identified that carers were undertaking tasks for which they were not primarily employed, even

if such interventions brought about desirable social benefits for the child and their family.

Provision of services for this group of children crossed all agencies and sectors; lack of visionary management and multi-agency working led to inefficiencies and wastage of resources, which in turn resulted in health and social disbenefits to children and their families. In particular, almost all children and young people attended a multitude of out-patient appointments to see doctors at various hospitals and clinics. There were financial and time costs to parents of making multiples of journeys to be reviewed by doctors in different hospitals and feelings of dissatisfaction that doctors did not have an overall view of the collaborative aims of clinical management which resulted in poor communication, duplication of services, and no overall plan of care. Children would benefit from multi-disciplinary clinics (such as the clinic set up in Bristol) whereby all the doctors and therapists involved in a child's care hold an annual joint clinic and develop a combined plan for each child.

Autonomy concerning the organisation and delivery of services was found to enhance feelings of self-respect and self-esteem in one family who managed their own independently financed care package. Almost all other families (particularly those with complex care packages) did not experience the same sense of control over the services they received. Although one other family had requested to manage their own care package (which had been resisted by doctors), almost all other families had not been provided with information concerning independent brokerage schemes.

Additional social benefits were derived from being able to take regular breaks and holidays. Breaks and holidays were associated with increased costs to parents if they had to pay to take their employed care team with them. There was insufficient provision of both in and out of home respite care for children dependent on ventilators, which conferred greater social disbenefits especially for mothers.

Flexible child-centred employed care teams

Most parents of children with complex ventilator-dependency required support from the state (and to a lesser extent from the voluntary sector) in the form of employed carers to manage their children at home (mean cost £57,407 per child per year; range £0 - £181,126; SD £63,710).

Costs were reduced for the state (but increased to parents) if unqualified carers were recruited. A well managed and trained team of nurses and/or carers who were flexible, accommodating and able to meet both the health and social needs of children and their families, was associated with increased social benefits in the form of positive life experiences and feelings of satisfaction for both children and their parents. Eighty-four percent of children with complex needs and employed carers had not gained maximum social benefit from their care team, due to unfilled shifts and inflexible or ineffective working practices (especially around risk management and coordination of care). Mothers in particular experienced social disbenefits and increased time costs if they were required to replace, back up, or supervise carers. Once home, professionals were concerned with reducing support costs, whereas parents were generally concerned with maintaining their current level of support and wanted more resources to address unmet needs (especially social).

Accessible, high quality housing

There was a high financial impact to parents (and in some cases the state) of providing an accessible and good quality home. Accurate figures were not available regarding the costs to parents generally, or the state specifically in terms of providing local authority accommodation or means tested disabled facilities grants of up to £20,000 per child.

A good quality, spacious and accessible home and garden was associated with considerable social benefits. These benefits included, privacy if the home was used as a place of employment, varied life experiences if the home was used to entertain and accommodate friends, and positive feelings of satisfaction for both children and their parents. Around 25% of families did not gain maximum social

benefit from their accommodation as they lived in poor quality houses that were inaccessible and lacked space for carers and storage. Lack of privacy from carers caused families to experience high levels of stress and dissatisfaction, which had a major negative impact on their wellbeing and overall quality of life.

The housing needs of disabled children have until recently been under-researched in terms of policy and practice. Housing issues have been given prominence by the publication of several studies carried out alongside the current programme of research (Heywood, 1996, 2001; Oldman and Beresford, 1999a,b Beresford and Oldman 2000, 2002; Bevan, 2002; McKeever, 2002). The current studies add to the evidence from these contemporaneous studies that a good quality home has the potential of serving 3 functions: to provide shelter; improve health outcomes, and facilitate social inclusion (Arblaster *et al.* 1998).

Guidance around the discharge management of ventilator-dependent children is one of the few documents to include a housing assessment as part of the discharge process (NHS Executive North West, 2000). Achieving the desirable outcome of an accessible home that brings about an improvement in health-quality of life of the child, however, remains elusive for some families.

Parents were, in the main, burdened with the financial responsibility of providing an accessible home for their disabled children; many parents wanted or needed financial help in order to realise the health and social benefits of their housing. All the aforementioned contemporaneous studies provide evidence that demand for local authority disabled facilities grants outstripped supply and the application process was overly bureaucratic and stressful to parents. Given the increasing numbers of children and young people who are dependent on medical technology, local authorities will need to consider the adequacy of their budgets (although this would be easier if accurate incidence and prevalence data were available).

Parents resented the means testing and wanted the test of resources to be based on the child's income, as was the case with disabled adults. The

increasing value of house prices has however clouded the issue of equity as parents have benefited financially from the award of grants to improve their homes.

The limitations on what the grant could be used for did not always bring about the intended improvement in the quality of children's lives. Consideration should be given to the requirements of children who have employed carers and would benefit from additional room to accommodate them. Introducing a small amount of discretion and flexibility to the criteria in specific circumstances could bring about a considerable improvement in the quality of lives of children and their families, for little additional cost.

Accessible and adapted domestic transport

There was a high financial impact on parents of providing accessible and adapted domestic transport. Accurate figures were not available regarding the costs to parents generally, or the state specifically in terms of financing Motability schemes. Twenty-four children received the mobility component of the disability living allowance.

Children under 3 years of age did not gain maximum social benefit from being able to get out and about as they were denied the mobility benefit. Around half of children experienced social exclusion as they could not get out and about as they would like; this engendered feelings of dissatisfaction, low self-esteem and stress.

Access to and appropriateness of education services

Eighteen ventilator-dependent children and young people required additional support in the classroom and/or transport to school (mean £7,143 per child per year).

A high quality education conferred additional social benefits for all children, especially around social inclusion, and opportunities for some children to enter higher education and employment. Around 40% of children did not gain

maximum educational or social benefit from their provision. Children missed school because of sickness, absence of their carers and classroom support workers, and hospital or therapy appointments. Not being able to gain optimal benefit from an education was a major source of dissatisfaction to parents and children alike.

Findings add to the body of evidence that active inclusion of disabled children in mainstream schools has major educational, social and economic benefits (Alderson and Goodey, 1998b; Alderson 1999). A good quality education has the potential to enable disabled children and young people to enter careers and follow life courses of their choosing (Ball, 1998). Although some children in the current studies reported good experiences, not all children received levels of support that enabled full inclusion and learning. It took a long time to undertake and process statements of educational need, and there were insufficient classroom assistants with experience in supporting ventilator-dependent children. The implication for local education authorities is that barriers that stifle access to resources still need to be removed and provision of classroom assistants needs to be increased to meet demand.

The procurement and delivery of equipment and disposable supplies

All children required medical devices and equipment costing on average £7,220 per child per year (range £1,894 - £20,186; SD £4,689). Children consumed varying amounts of disposable equipment and supplies (range £1,000 – £15,876) with an average cost of £8,133 per child per year.

Having the right equipment that was modern, portable, reliable, with a long battery life, that was easy to use and comfortable, conferred significant social benefits as children could get out of the home and be included in social activities. Forty-eight percent of children did not gain maximum social benefit as they required additional items of equipment; mainly powered wheelchairs, without which they were immobile, dependent and socially excluded. The voucher scheme for the provision of wheelchairs was inequitable and families turned to the voluntary sector to provide free resources.

Parents gained maximum social benefit when an efficient system was in place to order and deliver disposable equipment and supplies to their home. Sixty percent of parents reported significant disbenefits around time costs and dissatisfaction when systems failed. Children were also dissatisfied when vital supplies ran out.

Findings add to recent evaluations that also ascertained the provision of equipment (especially wheelchairs) needed to be organised more effectively in order to bring about the desired social benefits for children (Audit Commission, 2002a; Beresford 2003).

The supply and delivery of significant quantities of disposable products to domestic homes has not been planned for and families are suffering as a consequence of ad hoc arrangements. Families need product information and training, an efficient system of supply and distribution that takes into account the limited storage space in domestic homes, and access to help in an emergency. New logistical models of supply and distribution to domestic homes need to be developed and tested. Economies of scale may be achieved by developing joint supply and distribution networks with adult long-term ventilator users.

Findings from the current studies concur with the Audit Commission's recommendations, particularly around the need for strong leadership and efficient supply practices in the NHS (Audit Commission, 2002e). The Audit Commission found that only 14% of Trusts had fully integrated computer systems that could manage electronic requisitioning, ordering, invoice matching and payment. Many processes that were known to reduce procurement costs, such as consolidating orders and invoices, rationalising the supplier base and reducing the number of different products, were still not being taken up by many Trusts. The Audit Commission did not however consider the procurement, supply and delivery of equipment and disposable supplies to children's homes. There is a clear need for policies around procurement and supply to include the needs of high resource users such as ventilator-dependent children living in the community and for their service provision to be improved.

Access to and appropriateness of services for minority ethnic families

Twelve families in the current studies were from minority ethnic communities and received levels of support services that were commensurate with the range of costs for the entire group. Not being able to speak and/or read English conferred disadvantage in terms of understanding information that was only provided in English and being able to articulate the desired outcomes of care and services. A high proportion of employed care in the home was provided by nurses and carers from outside minority ethnic communities. This could be both a social benefit and disbenefit; mothers had the opportunity to learn English by speaking with English speaking carers, and carers took on a positive advocacy role in helping families to negotiate access to services. But by default, homes could be exposed to the different cultural norms and values of carers, which could cause tension and feelings of dissatisfaction. Children living away from their families for prolonged periods experienced multiple social disbenefits such as loss of their first language and assimilation into a culture other than their own.

Findings add to the growing body of evidence concerning the disadvantage that families from minority ethnic communities experience in relation to accessing and negotiating their way around services (for example, Fazil *et al.* 2002; Bywaters *et al.* 2003). Services did not always promote or reflect the needs of children from minority ethnic communities and had negative and unintended consequences; this situation is unsatisfactory and needs addressing.

'Looked after' children as a marginalised group

There was a high financial impact on the state of acting as a legal parent to 'looked after' children; total annual support costs £337,535 for one child (costs cannot be generalised to other 'looked after' children).

Children who were looked after by the state, especially those children from minority ethnic groups, or those living in hospitals, faced considerable social disbenefits that affected their overall quality of life and inclusion in society. In particular it was more difficult to gain access to these children and young people

in order to ascertain their views and opinions. 'Looked after' children experienced high levels of social exclusion and a lack of control over their lives (especially around prolonged hospitalisation), which engendered feelings of dissatisfaction and low self-esteem. 'Looked after' children were not always conferred their rights in law to independent advocacy and representation.

Findings also provide evidence on the substantial impact of having ambiguous legal and social status of having the state as your parent. The current studies add to the evidence that children looked after by the state have poorer outcomes generally (Morris, 1998a,b) and it is more difficult to access their views and experiences. In a contemporaneous study, Knight (1998) similarly found that children living away from their families did not always have access to an independent advocate as outlined in the Children Act (1989). There are too few trained advocates and this situation needs addressing so that children's rights in law are fulfilled.

Families of some 'looked after' children may be able to manage their children safely at home if additional qualified nursing support is provided, which will have cost implications. If, for whatever reason, children are not able to live with their birth families, it is not in the child's best interests to grow up in hospital whilst complex social problems are resolved. Family-based options such as foster care should be the preferred option to institutional care.

Three children in the current studies were looked after by the state and were not always aware of the reasons why. One child in foster care, whose mother had joint parental responsibility with social services, was not able to participate as he wanted because consent was not forthcoming from his mother. Findings concur with other studies that describe the high levels of risk and likely negative consequences for these children, and reinforce the need for child-centred policies that aim to promote the social inclusion of disabled and 'looked after' children.

Practicality of implementation

Findings from the 3 research themes relating to the costs and benefits of long-term ventilation provide evidence that help address some of the criteria, including organisational capacity, workforce implications and acceptability to children and their families. The policy drivers that act as a lever for change will be discussed in detail in section 9.3.

Organisational capacity

Demand for services, and in particular respite care services, for children who use complex medical technology was found to be far greater than supply. Almost all children with complex needs around ventilator-dependency spent prolonged periods of time in hospital due to insufficient capacity within the provision of community services. Some children accessed a children's hospice for respite care, which was highly valued but not available to all children. In particular provision was especially lacking for teenagers and young adults.

Workforce implications

Few non-parent carers were available who were trained and insured to provide this type of technical care. Due to shortages of appropriately trained carers, managers sought to restrict the roles of carers working in domestic homes to meeting the health needs of the child. There has been little forward planning concerning the numbers of non-parent carers who will be required to care for these children at home. Qualified nurses were found to increase the cost of care packages considerably and few NHS Trusts had rolling programmes in place to recruit and train sufficient numbers of unqualified carers specifically to care for ventilator-dependent children.

Acceptability to children, young people and their families

One of the well known difficulties that families of disabled children in the current study faced, was that the basic and costly needs of caring for their child, arose at precisely the time when they could least afford it. Seventy-six percent of families had a total household income of under £25,000 per year; 26% were lone parent families, 44% of families had no adult in employment and 56% of

families had debts (excluding mortgages) of up to £20,000 that were used to purchase vehicles and essential items. Families received additional welfare benefits (mean £7,380 per child per year; range £821- £15,334).

All families had an expectation that the state should provide additional financial support and services to meet the additional costs and burden of caring for their disabled children. Like in other studies around poverty and childhood disability, for most families, there was a mismatch between earnings and the needs of their child over their lifetime (Smyth and Robus, 1989). Lone parents did not benefit from child support payments from absent fathers; the financial stability of these families would improve if fathers contributed financially to the costs of bringing up their children.

An adequate income provided more of a cushion to cover the increased costs of caring for a disabled child and conferred social benefits such as varied opportunities to experience travel and purchase luxury goods. The financial stability of the family had a direct impact on their ability to provide a suitable home and accessible transport. Lone parent families in particular experienced financial and social disbenefits if their only source of income was derived from welfare benefits; a low income was associated with feelings of dissatisfaction and stress, and limited opportunities to enjoy a varied social life outside of the home (especially holidays and breaks).

Unlike in the current studies, Lindahl *et al.* (2003) reported that ventilator-dependent adults in Sweden did not mention money as a problem in a study exploring quality of life and home care. Lindahl *et al.* concluded that this was because the Swedish social insurance system provided patients with an adequate income; the replacement income paid in Sweden was higher than the equivalent welfare benefits in England.

Caring for ventilator-dependent children is associated with enormous time costs for parents (for example, Curren *et al.* 2001); for many families the time costs around caring were over and above those associated with disabled children generally. Many families were unable to care for their children at home without

employed care. The financial impact to families was made worse by some models of service delivery, whereby parents (principally mothers) were expected to replace, supervise and/or act as back-up to carers, which limited their opportunities to obtain paid employment and engage in social activities outside of the home. Whilst unqualified carers were found to be cheaper to the health service and sometimes more flexible than qualified nurses, their employment could result in many unintended consequences for families. Greater consideration needs to be given to the balance of qualified and unqualified employed carers when configuring the skill mix of teams to work in the home; potential cost savings to the NHS need to be balanced against the potential social disbenefits to parents. Relatively small adjustments to the balance and flexibility of qualified and unqualified carers within an employed care team could potentially have a positive impact on the financial stability and quality of life for families (especially mothers) at home.

Measurability of Implementation

Findings derived from the 3 research themes relating to costs and benefits of long-term ventilation provide evidence to address the criteria concerning the robustness of monitoring systems and the practicality of setting up new monitoring arrangements.

Robustness of existing monitoring arrangements, including indicators for both process and outcome

Synthesis of findings across studies found that few services were monitored to ascertain whether the services provided brought about desired outcomes from the perspective of the child and their family. Examples were found of children who had very expensive care packages, but the ineffective and inflexible manner in which services were organised and delivered had a profound and negative impact on the quality of life of the children and their entire families.

The Local Government Act (1999) requires public services to be delivered in accordance with 'Best Value' principles. All services must be subject to continuous improvement in the way in which its functions are exercised, having

regard to a combination of economy, efficiency and effectiveness. In the current studies, Best Value performance indicators tended to focus on processes and inputs as opposed to impacts or outcomes. Providing a package of support services to enable the child to live at home was often seen by professionals as a means to an end in order to free up a much needed intensive care or hospital bed. Few packages of support were evaluated to see if the services and resources provided brought about the desired health and social outcomes for the child and their family. Review of care packages was also dominated by a need to save money whenever possible.

The arrangements for the statutory monitoring and inspection of the multi-agency services that contributed to a child's overall package of care were also found to be inadequate. Not all children and young people were monitored in accordance with guidance outlined in the Children Act (1989) (especially children living in hospital) and this requires addressing. Ten inspection agencies were involved in auditing the quality of services provided to children in the current studies, including:

- Commission for Health Improvement;
- Commission for Social Care Inspection;
- Social Services Inspectorate;
- National Care Standards Commission;
- Commission for Health and Audit Inspections;
- Magistrates' Courts Service Inspectorate;
- Office for Standards in Education;
- Adult Learning Inspectorate;
- The Audit Commission, and
- The Office of the Deputy Prime Minister.

All of these agencies used a different framework for inspecting services and in the main inspections were conducted in isolation. Some children in the current study also slipped through the inspection net; hospital wards were not classified as residential units, and thus were not visited by the Social Services

Inspectorate, who were ideally placed to identify that children's social and developmental needs were not being met in such environments.

Practicality of setting up new monitoring arrangements

The Green Paper *Every Child Matters* (DoH, 2003b) aims to address these anomalies in the inspection process. All inspectorates must work together to develop a single multi-agency inspection framework for children's services that will be introduced in 2005. The inspection methodology will be matched against the standards in the National Service Framework for Children (DoH, 2003a). This integrated system of monitoring services is broadly supported by findings from the current studies and would be welcomed by parents and children.

Summary of contribution of findings to the evidence base

In summary, the current studies have contributed new and original evidence demonstrating:

- The need for greater service provision as numbers of ventilator-dependent children are increasing steadily;
- Increasing prevalence of the risk factors for developing ventilator-dependency;
- The scope for health benefit in most but not all applications of long-term ventilation;
- The impact of ventilator-dependency on affected children and their families;
- The cost to the NHS, social services (excluding housing provision) and education authorities;
- Areas where existing services are not meeting needs or where there is variation in the quality of services provided;
- The inability of the NHS and social services to meet the needs of 'looked after' children and provide appropriate services for minority ethnic children;
- The effectiveness (or not) of service models;

- The practicality of implementation in terms of: the lack of organisational capacity within the NHS; workforce implications; acceptability of models of care from the perspective of children and their families, and
- The lack of robustness of existing monitoring arrangements, including the need to identify indicators for assessing needs, processes and outcomes.

Significant gaps remain however in the evidence base, including:

- The actual numbers of children and young people affected;
- Accurate estimates around the predicted increase in need;
- The potential relationship between numbers of intensive care beds and increased prevalence;
- The costs to the individual and society;
- The magnitude of benefit for all groups of children and young people;
- The health and social needs of children and their families;
- The practicality of implementation in terms of increasing organisational capacity within the NHS and the actual costs of implementation, and
- The efficacy of models for monitoring quality and outcomes of care.

The need for a more extensive evidence base and future research directions to fill the gaps in evidence identified during this programme of research will be outlined in Chapter 10.

In the next section of this chapter, the ongoing requirement to define the needs and outcomes of care with ventilator-dependent children and their families is explored in greater depth.

9.2 Defining needs and outcomes of care

One of the major conceptual issues that cut across all 3 themes concerning the costs and benefits of long-term ventilation in children was the lack of clarity regarding the health and social needs of children and their families, and the outcomes of the care and services provided. The ongoing requirement to

define needs and outcomes of care is important, and has practical application in terms of implementing policies and in particular the commissioning and monitoring of services.

The relatively low prevalence and slow but increasing trend in the incidence of this new population, helps to explain why this group of children were not recognised as requiring services for their specific needs until the population reached a critical mass in the mid 1990s. As a consequence the responsiveness of health, social and education services has been extremely slow. State-funded provision has developed in an ad hoc manner in response to the blocking of intensive care beds by ventilator-dependent children. The voluntary sector and charities have been quicker to develop respite services for ventilator-dependent children, but supply has been insufficient to cope with demand.

The joint framework used by agencies for the assessment of need in children and their families focuses on the child in question and does not include the needs of siblings or the wider family (DoH, DfEE, HO, 2000). The current studies found that this approach to the assessment of need had a number of unintended impacts, as existing services did not meet the holistic needs of over half of the children with complex needs around ventilator-dependency, and their families. Health services tended to focus on health needs and social services tended to focus on social needs, whereas families wanted a generic person who could deal with both aspects of need. Many care packages were designed primarily to meet the health needs of ventilator-dependent children and parents wanted their entire family unit to be included in the assessment of need. There was also great variation in services provided in terms of ease of access, user and carer experience, quality of services provided, and patient outcomes.

Review of the literature revealed little evidence concerning the health and social needs of ventilator-dependent specifically, or disabled children generally, to inform the commissioning of services, or models of service delivery. One notable exception was a contemporaneous study carried out by Turnbull *et al.* (2000), who like the current studies found that both the health and social needs

of disabled children and their families needed to be addressed before families benefited from an improved quality of life.

Little consultation concerning children's needs and desired outcomes of care has been undertaken with children and their families. In the absence of an evidence base and effective procedures around defining and monitoring needs and outcomes, parents have used complaining as a way of getting things done. This situation is unsatisfactory and needs addressing. More attention needs to be paid to defining and evaluating outcomes of care to ensure that services are tailored around families (as opposed to the other way round), and that services offer value for money.

The Department of Health and the Department for Education and Science have recognised that children's views need to inform policy-making and practice (Lightfoot and Sloper, 2002). In a joint initiative launched in 2003, they have pledged to listen, hear and respond to children's views regarding their care and treatment (DoH, 1998c, 2002c, 2003c). Findings from the current studies support the need for such an initiative and children would welcome more involvement in both policy and decision-making.

Current methods of outcome assessment in health services research are dominated by quantitative measures; findings indicate however, that generic measures will only be useful if the questions asked are relevant to the service aims, and meaningful to children and young people. Qualitative findings in particular alluded to the outcomes that children and their families wanted, but they were not necessarily the intended outcomes of the health services provided (for example, support with housework, and experience of a good social life by the entire family).

It may be that parents and children are alluding to the life that they would like, rather than the life that services limited by finite resources can conceivably provide. The main adult (ventilator-dependent) role models cited by children in the current study were Christopher Reeve and Stephen Hawking; both are well

off economically and can afford a lifestyle beyond the reach of most ventilator-dependent children (Reeve, 1998).

There is a very small literature concerning the social outcomes of care and services for disabled children and those with complex healthcare needs (SPRU, 2000 a,b,c). Similar to disabled children generally, for ventilator-dependent children and their families, quality of life was firmly associated with both health and social inequalities (Rogerson, 1995). Factors such as stress were found to have a direct impact on the quality of their life experience (Brunner, 1997). Packages of support services were not primarily designed to deal with economic, environmental and psychological stress as experienced by the families of ventilator-dependent children.

Findings add to understanding concerning the impact of social support and quality of life (Helgeson, 2003), the long-term impact of caring on maternal health (Thyen *et al.* 1998), and the social exclusion experienced by the entire family (Cohen, 1999). Relatively small adjustments to the roles of nurses and carers, in particular around incorporating social needs into their remit, combined with multi-agency working, may bring about the desired social outcomes for children and their families. The use of a generic carer with both a health and social focus may be a more appropriate person to work with families in domestic settings.

The Department of Health is currently funding a research programme to develop outcomes of care with children who have complex healthcare needs, with the aim of establishing the effectiveness of health and social care interventions (SPRU, 2000 a,b,c). In combination, the Best Value initiatives and research into outcomes offer an opportunity to use the evidence around costs and outcomes of care derived from the current studies to develop effective services, that bring about the desired outcomes from the perspective of children and their families. The current studies demonstrated that investment in child-centred, well coordinated and effective support services could help to minimise negative impacts and bring about the desired health and social outcomes from the perspective of the child and their family.

The final section of this chapter focuses on reviewing findings within a health and social policy context.

9.3 Review of findings in a health and social policy context

Since the programme of research reported in this thesis commenced, major policy reforms have been implemented by the Labour Government. Common political objectives underpinning reforms have been to maximise a combination of the quantity and quality of the lives of people, reduce inequality and social exclusion, whilst at the same time demonstrating prudent management of the economy.

The major national policy drivers that are pertinent to the current studies include reforms of the health service including the NHS plan (DoH, 2000a), which sets out the investments and reforms that aim to transform the NHS into a patient-centred service. In relation to child policy, the national drivers include the National Service Framework for Children to reform child health services (DoH, 2003a), and the Quality Protects Programme to reform social services (DoH, 1998a,b). A Green Paper (*Every Child Matters*) is currently undergoing consultation (DoH, 2003b). In addition, elements of child policy are integrated throughout policies that cross Government departments (for example, disability legislation). A Minister for Children has been appointed to chair a Children's Taskforce and is charged with ensuring that the cross government agenda is delivered for children.

Findings from the current studies are pertinent to specific areas of health, social and child policy reforms including:

- Social exclusion;
- Child poverty;
- Integration of services across sectors;
- Direct payment scheme;
- Workforce planning, and
- Funding of services.

These headings will be used as a structure to review findings within a policy context.

Social exclusion

Long-term ventilation is now a routine and high cost intervention that confers health benefits to most children, but without focused and continuing support to meet both the health and social needs of the entire family, long-term ventilation can confer considerable social disbenefits, especially social exclusion.

Ventilator-dependent children experienced social exclusion for a host of reasons such as: living in unsuitable homes, being unable to access transport, being excluded from buildings and activities, frequent absences from school; unmet need for, or poor quality equipment; being unable to communicate effectively; living in poor or financially unstable families, and having few opportunities to make friends and play.

The reduction of social exclusion and the welfare of disabled and 'looked after' children are central to the Government agenda. A raft of policy initiatives that span Government departments were rolled out during this programme of research. These include:

- The *Quality Protects Programme* aims to ensure that the needs of 'looked after' and disabled children are adequately assessed and met, enabling them to live with their families or other appropriate settings in the community (DoH, 1998 a,b);
- The Green Paper *Every Child Matters* aims to ensure that every child reaches their potential through targeted services and child protection policies (DoH, 2003b);
- The *Disability Discrimination Act* (1995) includes the duty to adopt a strategic approach, in partnership with local users, towards more accessible play and leisure services for disabled children;

- Part III of the Act puts duties on providers of goods and services, including, from 2004, to remove barriers creating difficulties in accessing the physical environment of a setting;
- The Office of the Deputy Prime Minister is issuing practical guidance on accessible play;
- The Department for Culture, Media and Sport has launched *A Framework for Action on Disability*, including a commitment to enhance access to cultural and sporting opportunities for disabled children (DfCMS, 2003);
- The National Service Framework for Children will provide standards for services to apply nationally and locally to promote social inclusion, including standards for equipment services, housing and adaptations, and special needs transport. In particular, targets have been set to increase uptake of a Carers Special Grant by 2006 and improve the life chances of all children;
- The *Integrated Community Equipment Services Initiative* has set a framework for the integration and improvement of community equipment and mobility services enabling disabled children to get out and about with greater ease (DoH, 2001a);
- The Modernisation Agency has established a collaborative programme to support improvement in wheelchair services for disabled children;
- The Department for Education and Science (DfES) Pathfinder scheme supports initiatives to improve transport services for disabled young people;
- The Special Education Needs and Disability Act (2001), the Special Education Needs Code of Practice and the forthcoming Special Educational Needs Action Programme (DfES, 2003); all promote the inclusion of disabled children, and together with
- The White Paper *Valuing People* sets out an expectation that all local services will introduce person-centred planning for all young people moving from children's to adult services. It also requires Learning Disability Partnership Boards to identify a member with lead responsibility for transition issues, who will work with the Connexions

service to ensure effective joint working and continued inclusion of the disabled child into adulthood (DoH, 2001b).

All of these cross Government initiatives are consistent with the needs identified in the current studies and would be welcomed by ventilator-dependent children and their families as long as these policies translate into tangible services and positive outcomes that make a real difference to children's lives. None of these initiatives however are likely to make a real difference to the inclusion of ventilator-dependent children in society unless issues around risk management are addressed.

The issue of risk management poses a considerable dilemma. Findings illuminated different perspectives around risk:

- Doctors and nurses took actions designed to minimise risk, and were especially worried about untoward incidents such as ventilator disconnection and death;
- Children and young people wanted less surveillance and to experience more of life;
- Parents had varying opinions ranging from agreeing with their children to wanting more surveillance and some sued when things went wrong, and
- Managers wanted to minimise the potential for being sued and managed risk by eliminating them.

Given the current propensity of parents to sue for damages when untoward events occur, and the high incidence of accidental death in highly ventilator-dependent children (Janowski, 1984), the nervousness of professionals around risk management appears justified (Gilgoff and Helgren, 1992). There is however evidence in the literature that ventilator-dependent children can live relatively safely outside of hospital environments (For example, Ambrosio *et al.* 1998). Findings suggest that a more flexible policy is needed to replace the current rigid risk-averse approach, which aims to reduce risk by eliminating activities perceived to be 'risky'. When parents and young people are fully

conversant of the risks and are willing to accept them in return for an increased quality of life, then policies ought to be able to accommodate this position.

Child poverty

Three quarters of families with ventilator-dependent children were poor (annual income below national average wage) and impoverished further by having to provide for a disabled child.

The Chancellor aims to eradicate child poverty by redistributing wealth to poor families with children through targeted welfare benefits (some specifically for disabled children and their families) and favourable tax breaks (HM Treasury, 2000). This will provide families with more disposable income to increase their living standards and participate more fully in society.

Whilst it is unlikely that levels of welfare will increase substantially or match those paid in Scandinavian countries, a small amount of additional flexibility could potentially benefit the financial stability of families. In particular, children and their families were ineligible for some welfare benefits by virtue of living in hospital for prolonged periods; despite the fact they had unmet needs around social inclusion, their parents worked as unpaid carers and experienced increased costs at the same time as their incomes and opportunities for employment reduced or ceased. Likewise, children under 3 years of age were not eligible for the mobility component of the disability living allowance, despite the fact many children needed adapted transport to get out and about.

Integration of services across sectors

The current studies found that poor management practices and lack of a key worker with sufficient authority prolonged the hospitalisation of these children, and resulted in inefficient ongoing management of care packages in the community. These findings add to the wealth of evidence relating to the positive impact of key working on managing complex packages of care. It is disappointing that this evidence has not always translated into local policies and procedures.

At the heart of the Government's modernisation agenda is a fundamental change in philosophy about how health and social services should be delivered. This involves a cultural change in the way in which services are designed and delivered around the children using the services and not the organisations delivering the services (DoH, 1998d). There has been a raft of cross-department policy developments to bring about multi-agency working and integration within disabled children's services generally. All are aimed at providing seamless services that start from user need and that involve children and young people who are dependent on ventilators and their families. Findings from the current studies broadly support the aims of these initiatives, which include:

- *Together from the Start* which provides a framework for multi-agency working for younger children (DfES and DoH, 2003a);
- *The Sure Start* programme, developed between the Department for Education and Skills and the Department for Work and Pensions, aims to deliver the best start in life by bringing together early education, childcare and health and family support under one umbrella;
- *The National Service Framework for Children* which will provide standards for services to apply nationally and locally;
- *The Quality Protects Programme* provides a framework and standards for multi-agency working;
- The Government's programme for extended schools, will offer opportunities for schools to act as the focal point for a range of services geared towards the needs of children and their families (DfES, 2003);
- The flexibilities incorporated into the Health Act (1999) will provide opportunities to pool budgets across health and social services;
- Children's Pathfinder Trusts aim to provide integrated health and social services;
- Children and Young People's Framework for Partnerships in Wales are responsible for overall planning of services for children and young people in each local authority (National Assembly for Wales, 2000, DoH 2003), and

- The appointment of a Children's Taskforce in England and Children's Commissioners in England, Scotland and Wales aims to highlight the needs of children across Government departments.

There has always been a danger that the needs of this heterogeneous low volume high cost group of ventilator-dependent children and young people may not have been recognised when major structural or organisational changes to health and social services were implemented. However, evidence from the current and other relevant studies was submitted to the National Service Framework for Children external working groups and collated in a report written by Barnardos (Ludvigsen and Morrison, 2003). The Department of Health subsequently commissioned a project to produce national clinical guidance on the management of ventilator-dependent children to supplement the National Service Framework for children. In particular, the guidance will include cost estimates and outcomes from this programme of research. This is an example of research findings having an influence on evolving clinical practice.

Findings reported here imply that, apart from issues around discharge from hospital, the cross Government reforms to promote multi-agency working have had a positive impact on children and young people with simple ventilator-dependency and around half of those with complex ventilator-dependency. Those children with the most complex needs and whose resource use was highest have yet to reap the benefit of multi-agency collaboration, key working and improved communication across sectors. In some areas, specific barriers to 'policy transfer' have stifled organisational change and these barriers need to be removed before these policies can be realised in practice.

The aims of Government reforms also appear visionary regarding the concept of seamless, and high quality services that meet children's global needs. Experiences of some families fell short of this vision, in that many children and young people received disjointed, over-stretched and in some cases low quality services. In particular, there did not appear to be a seamless service between hospitals and the community; families described the interface as more like a black hole.

The aspirational vision of current policy initiatives, however, was welcomed by children and their parents, together with whatever actions were required to implement changes; especially by the parents who currently had or wanted primary responsibility for coordinating their own services and care packages.

Direct Payment scheme

Under the 1996 Community Care (Direct Payments) Act, social services departments are empowered to make cash payments to service users in lieu of direct service provision (DoH 1998e). Making use of the direct payments development fund may allow children to get more individualised support and so facilitate greater social inclusion (DoH, 2000c). Only one family in the current studies had been offered an independent brokerage scheme and benefited from being in control of their lives. The prospect of handling an annual budget of £104,000 upwards (and in one case, £331,619) may be an added stress rather than liberating to parents. Nonetheless some parents may feel more in control and satisfied if they managed aspects of their care package (such as the purchase of respite care) to suit their needs.

Health and social care professionals need to be fully conversant with such schemes so that parents and young people can make informed choices about the management of their packages of support services. The current studies, however, indicate that agencies do not always actively promote such schemes. For example, a wealth of information for patients (including a video) is available concerning the direct payment scheme, but only one family in the current studies had seen it (DoH, 1998e, 1999b, 2000d).

Workforce planning

The recruitment and retention of appropriately trained staff was found to be key in terms of providing effective services. Nationally, the NHS has suffered from significant problems with the recruitment and retention of staff. During the course of this programme of research a number of policy initiatives around recruitment and retention were rolled out, including:

- The Department of Health has set specific targets for recruitment of staff and invested in additional training places for doctors, nurses and professionals allied to medicine;
- The Health Act (1999) includes flexibilities that allow agencies to make jointly funded appointments;
- The Green Paper *Every Child Matters* (DoH, 2003b) provides specific guidance on the issues involved in staffing children's services;
- The National Service Framework for Children (DoH, 2003a) makes recommendations on recruitment and retention of staff, and
- The Audit Commission Report *Recruitment and Retention – A Public Service Workforce for the 21st Century* provides guidance on the recruitment, retention and development of staff skills (Audit Commission, 2002d).

Whilst these initiatives are welcome, it has yet to be seen if these policy initiatives will translate into more carers and nurses with specific training to care for children and young people dependent on ventilators. This type of 'high-tech' care in domestic settings was found to be not particularly attractive to qualified nurses and more incentives may have to be built into the system. Some parents were also resistant to having unqualified carers in their homes, even though they were said to be easier to recruit and retain than qualified nurses.

The current studies also found that for children with the most complex needs, a multitude of professionals visited the family home, each to deliver a small but important aspect of the overall care package. More thought needs to be put into developing new ways of working in order to improve continuity of care and reduce the number of professionals involved. None of the cross department strategies concerning human resource issues looks specifically at the need to consolidate roles across professions or the possibility of training a new generic health and social carer with the appropriate technological skills.

Funding of services

The present system of awarding block contracts for intensive care services was found to act as a disincentive to transferring children home, as additional resources were required to purchase a package of community support services where they had not existed before. Current budgets to purchase packages of community support services were found to be inadequate to meet increasing demand. As a result, paediatric intensive care provision was still being used inappropriately to house ventilator-dependent children in the medium to long-term thereby depriving other critically sick children of their best chance of survival. These are the same conditions that led to a public inquiry when insufficient intensive care beds were available due to a high percentage being occupied by ventilator-dependent children (Ashworth, 1996). If rigorous estimates of the incidence and prevalence of children dependent on ventilators were available then a more appropriate sum of money could be allocated to purchase packages of support services to enable children to live at home, thereby freeing vital intensive care beds.

The population of children dependent on ventilators is relatively small and not yet enumerated, but individual children have the ability to consume very high levels of resources. Having estimated the costs of care for ventilator-dependent children it became clear that individual Primary Care Trusts might have insufficient funds to manage a significant number of these extremely high resource use children. In most areas, groups of Primary Care Trusts have subsequently formed regional children's intensive care consortia to commission services for high resource use children such as those dependent on ventilators. According to the doctors who participated in the current studies, demand for services still outstrips supply and children are still waiting long periods of time in hospital awaiting discharge. Central funding from Government may be required to support small groups of people who consume disproportionately high levels of resources (Barker, 1996).

For the reforms to work, accounting responsibility needs to be clearly established; ideally money needs to follow the child to purchase essential

equipment and services and should not be seen as the sole responsibility of the service who originally identified the need. The Government, however, has yet to establish a detailed activity-based costing system that determines the accounting responsibilities for the substantial cost drivers (such as the cost of a ventilator or the funding of full-time carers). It has yet to be seen if a seamless service can be achieved in any system where competing and contested decisions concerning money ultimately determine a package of care to enable a child to live at home.

The use of NHS vouchers to purchase private sector services

Findings from the current studies indicate that the voucher scheme initiative that aimed to give wheelchair users more choice and financial aid from the NHS if they chose to purchase a wheelchair provided by the private sector, had a number of limitations and unintended consequences. Primarily, those families who used a voucher from the NHS to purchase a more expensive chair from the private sector found that they could not always afford to pay the high cost of servicing and maintenance. These families were on relatively low incomes and were not fully aware of the potential ongoing costs involved. For most families, the voucher scheme was not an option as they managed on low incomes and did not have the additional money required to purchase a more expensive (and in their eyes a more desirable chair) from the private sector. Some children who had been compensated as the result of accidents said they took advantage of the voucher scheme to purchase a more expensive chair. An evaluation of the voucher scheme initiative in 2000 also found evidence that some users who would otherwise have purchased a wheelchair privately had come to the NHS to take advantage of the voucher funding (NHS Executive and DoH, 2000). The report recommended that better value for money would probably have been obtained if the considerable funds that had been invested in setting up and managing the scheme had been used to improve the range and quality of wheelchairs from core NHS wheelchair services.

The voucher scheme initiative for wheelchairs is not dissimilar to current Conservative Party policy concerning the management of healthcare – namely ‘Patient Passports’ (Conservative Party, 2003). Indeed, it was the previous

Conservative Government who introduced the wheelchair voucher initiative in 1996. Findings add to the evidence that such voucher and passport schemes subsidise better off families at the expense of those less well off families who rely on core NHS services.

Long-term sustainability of service provision

In every healthcare system, resources are always scarce relative to the wants and needs of patients and although the current Labour Government have increased the amount of spending on the NHS and social services, this has to be balanced against increasing demand for services.

The most critical resource that enabled children dependent on ventilators to enjoy good quality of life at home was money that was efficiently spent and mainly redistributed by the state (Nosek and Holmes, 1996). There is evidence from the United States that politicians and citizens alike are reassessing how much they are willing to pay for health services for those with medical needs and few resources (Orlowski, 1993). Similarly, Hammer (2000) reported that the negative perceptions expressed by the local community around the ethics of funding long-term ventilation in Switzerland were one of the main problems experienced by families with ventilator-dependent children at home. Evidence in the literature also indicated that some UK taxpayers were not willing to pay for a substantial redistribution of resources and wealth to children who were dependent on ventilators; despite the fact that evidence from the current studies indicate that cost-effective (albeit very costly) packages of support services can potentially bring about real social benefits such as social inclusion (Hadden, 1996). Public attitudes to dependency and the welfare state can act as an arbiter of public policy; political parties can and do capitalise on peoples' fears when the tax system is perceived to be unjustly burdensome on those in work (Bradshaw and Mayhew, 2004).

The current studies found that demand for community services by children dependent on ventilators was outpacing year-on-year budget increases. Without substantial additional increases in spending on community services, the logical conclusion would be to fund packages of support services by making efficiency

savings. Efficiencies could be made by better management and more appropriate models of service provision, but it is unlikely that these efficiencies would be sufficient if there was an increase in the incidence of ventilator-dependent children or a significant reduction in Government spending on health and social services due to a downturn in the economy or a reduction in taxation.

In a national health service that is funded by taxation, revenue will depend on the level of taxation and the state of the domestic economy. Inevitably revenue will rise and fall in real terms in relation to these factors. The economic evaluation reported in this thesis was undertaken during a time when the UK economy was strong and substantial increases in funding had been invested into children's health and social services. Despite these increases in funding, demand for services still outstripped supply as numbers of children and young people dependent on ventilators and other medical technologies continued to grow.

There are many historical examples whereby financial pressures and economic recession have forced the reversal of aspirational plans for the provision and delivery of services that have raised public expectation. Beecham and Knapp (1992) cite the example of the policy to provide community services for long-stay psychiatric patients whereby the needs-led intentions around the commissioning of services have given way to supply-led services that are driven by constrained budgets and the latest administrative or legal ruling regarding eligibility.

Given that the 35 children and young people dependent on ventilators in the current economic study consumed resources costing over £6.2 million in the previous 12 months, an economic downturn or political imperative to decrease spending on health and social services would undoubtedly result in cuts to their service provision. If the reduction in service provision for home care was severe, some parents may not be able to cope without additional employed carers. In the United States, when the costs of care for ventilator-dependent children started escalating in the early 1990s, benefits were cut and parental contributions increased. Many states passed legislation to ensure that patients

dependent on ventilators were cared for in the cheapest institutional facility irrespective of the negative psychosocial outcomes to the patient. Likewise, low and middle-income countries have prioritised their spending on public health programmes and therapeutic interventions to benefit greater numbers of children.

9.4 Summary

This chapter brought together the results of the research reported in this thesis within an evidence based and policy context.

When matched against criteria for framing evidence to underpin the commissioning of services, synthesised findings concerning the costs and benefits of long-term ventilation provided an indication as to the adequacy of the current evidence base. Findings from the current programme of research were found to have made a substantial contribution to the evidence base, but gaps remained in several areas, principally around incidence and prevalence, the costs to society, the magnitude of benefit for all children, the needs of children and outcomes of care, and the most effective ways to deliver services.

The synthesis of evidence identified the requirement to define the health and social needs, and outcomes of care, as one of the most important gaps in the evidence base. This issue was considered particularly important, as it has practical application in terms of the commissioning of services based on needs, costs and outcomes. Two factors were found to have contributed to this situation, including the inflexibility of the joint framework for the assessment of need in children which did not include the wider family, and the lack of research into the needs and outcomes of this group. Further research needs to be undertaken to establish what children and their families want from the care and services provided.

Finally, synthesised findings were then reviewed in the light of current health and social policy initiatives. Here a complex picture of major national and cross-departmental policy initiatives emerged both in relation to health and

social policy and child policy. These numerous policy initiatives aimed to transform health and social services into child-centred services, promote the inclusion of children (especially disabled children) in society and eradicate child poverty. The aims of these initiatives were found to be broadly in line with findings.

The next chapter concludes the thesis.

Chapter 10

Conclusions

10.0 Introduction

The thesis concludes with a final chapter that outlines future research directions and makes the case for a more extensive evidence base concerning small volume high cost groups, such as ventilator-dependent children. Finally, the thesis closes by exploring the unresolved dilemma as to what level of resources, and which health and social outcomes, the state should reasonably be expected to meet. This unresolved dilemma is examined in the light of legal imperatives versus evidence based medicine.

10.1 Future research directions

Consistent with the presentation of findings in Chapter 9, future research directions will be matched against relevant National Service Framework criteria for framing evidence around health problems and medical conditions, including:

- Demonstrable need and predicted increase in need;
- Magnitude of benefit;
- Effectiveness and cost-effectiveness of interventions, and
- Effectiveness of service models for implementing interventions.

Demonstrable need and predicted increase in need

The priority of future research has to be the need to enumerate the size of the population and predict prevalence and incidence in order to plan appropriate service provision.

The Department of Health is currently rolling out an Information Management Strategy that aims to inject significant investment into information technology for

the health service (DoH, 2002e). The new Government Information strategy aims to record and codify each patient journey through primary and secondary care. This strategy offers a unique opportunity to enumerate the population of children dependent on ventilators, calculate their resource use, and identify the relationship between the increasing provision of intensive care beds and numbers of children affected. But this will only happen if individual ventilator-dependent children can be identified from routine ICD codes and the system is strengthened to ensure this happens.

In addition, the British Paediatric Surveillance Unit could be funded to undertake a prospective nationwide survey of new cases of children dependent on ventilators in order to provide accurate estimates of incidence over a defined period (Hall and Nicoll, 1998).

Finally, exploratory fieldwork in Chapter 2 identified 5 reasons/factors underpinning the emergence of ventilation-dependent children. Additional research is needed that utilises a more rigorous theoretical framework in order to replicate these findings.

Magnitude of benefit

Further research needs to be undertaken to distinguish groups of children and young people for whom the utility of long-term ventilation is highly beneficial from those for whom long-term ventilation is highly questionable. Further methodological development around the assessment of health-related quality of life is needed; findings from subsequent rigorously conducted studies may assist in the development of morally defensible criteria concerning treatment decisions and the allocation of finite resources.

In particular, one of the most challenging aspects of this programme of research was attempting to describe or measure the health-related quality of life of children and young people with profound sensory impairments. Both qualitative and quantitative methods were found to be inadequate.

The difficult question with this group of children with profound sensory impairments is around the interpretation of the costs versus the perceived benefits of treatment. In the current study, all parents of children with sensory impairments said that their children were happy, well cared for and benefited from long-term ventilation (although many wanted more services such as respite care).

There is no obligation on doctors to give treatment which is considered to be futile and burdensome, especially when resources are finite and insufficient to treat all children. However, in high-income countries there is strong evidence that children with profound sensory impairments are being routinely treated with long-term ventilation as an intervention. Kaufman (2003) in her recent ethnographic study of long-term ventilation units in the United States described these units as *'new cultural forms that enable beings who are neither fully alive, biologically dead, nor 'naturally' self-regulating, yet who are sustained by modern medical practices, to exist'*. Kaufman concluded that the mere existence of such specialised units created a mandate for ensuring the maintenance of life. Further research is needed to determine the health-related quality of life of children and young people with profound sensory impairments and to explore in greater depth the decision-making processes of both parents and doctors.

Only qualitative methods were able to elicit an accurate picture concerning children's perceptions around their health and quality of life. Quantitative instruments that measured functional attributes (such as being able to walk) were not considered important by all children. Further research is needed to develop more appropriate child-centred ways of describing health and quality of life in children with physical impairments that can be used to inform decision-making.

The measurement of health-related quality of life was used as a discriminatory index to measure differences between a sample of ventilator-dependent children and a normative population of school children. Review of the literature found that no research had been undertaken to assess or classify ventilator-

dependent children in order to provide appropriate therapeutic interventions, or to evaluate the impact of different treatments or interventions over time. Further research needs to be undertaken to determine if health-related quality of life instruments can reliably assess the need for, and measure the outcomes of, treatment over time.

In terms of measuring the magnitude of benefit, findings challenged the assumption that lack of concordance between the scores of parents and children equated to an inadequate measure of health-related quality of life. Subtle gender differences were also found between boys and girls, and parents of boys and girls. More research with greater numbers of ventilator-dependent children needs to be undertaken to investigate whether concordance is greater on some domains than others; whether parents rate their child's quality of life to be better or poorer than their child's own ratings and to what extent scores are affected by the child's age, gender and illness status.

Effectiveness and cost-effectiveness of interventions

Further mixed-method research is required to develop a common framework for understanding outcomes of health and social care provided for children and young people who are dependent on ventilators. Such research may help to narrow the current gulf between the perceived needs of children and their families and the seemingly different aims of the services provided.

The current studies provide cost estimates primarily from the perspective of the NHS and education authorities. Further research is needed to provide accurate cost estimates from a societal perspective, including the contribution of parents and the costs of housing and transport provided or subsidised by social services and local authorities. If accurate data were available on incidence and prevalence, then the costs of supporting this sample of ventilator-dependent children and young adults could be used to estimate costs of caring for the entire population of young ventilator-dependent people in the United Kingdom. This is an important issue as commissioners and managers need to allocate an

appropriate sum of money to purchase packages of support services to enable children to live at home, thereby freeing vital intensive care beds.

Effectiveness of service models for implementing interventions

Current Government policies have not always brought about the desired changes in service delivery, even where rigorous evidence exists concerning the effectiveness of service models, such as multi-agency working. A process evaluation is needed to identify the barriers that prevent 'policy transfer' from evidence to practice so that reforms bring about maximum benefit to children and their families.

The evaluation of NHS provision found that this small group of children and young people consumed very high volumes of resources, there were significant inefficiencies in terms of costs, and around half of the patients were dissatisfied. At a Government level there is a widespread assumption that the private sector can provide services more cheaply and the development of public-private collaborations in the delivery of public services is a central strand in current Government policy in England (DoH, 2000ab, 2002b). Health services for children dependent on other technologies, such as home dialysis for children with renal failure, have for many years been contracted out to the companies who produce the technology (For example Baxter plc and Gambro plc). These private companies provide a range of integrated patient and product support services for children at home anywhere in the UK. Unlike with long-term ventilation, there have been few anecdotal or published reports of hospital beds being blocked due to insufficient community services, or levels of parent dissatisfaction with services expressed on a scale anywhere near that of families with children dependent on ventilators.

In the light of seemingly insurmountable barriers to improving the quality of NHS services, 8 NHS Trusts have recently entered into a public-private collaboration with the same private company to discharge and provide ongoing care management to ventilator-dependent children and young adults. Review of the literature revealed that there is little rigorously conducted research into the

outcomes of NHS public-private collaborations for children and their families generally. An economic study and process evaluation is required to assess the costs and benefits of private sector involvement in the delivery of services to ventilator-dependent children and their families. If the delivery of services by the private sector are found to be effective and cost-effective then the NHS will have more evidence to underpin the commissioning of further public-private collaborations.

Getting the children home from hospital within a reasonable length of time is a desirable and yet ill-achieved outcome of care. There is some evidence that discharge coordinators have a positive impact on reducing the time and costs associated with discharging children home. A randomised controlled trial could be used to establish the effectiveness and cost-effectiveness of hospital discharge coordinators to facilitate the discharge home of ventilator-dependent children. If found to be effective, this model could be replicated throughout the UK.

10.2 The need for a more extensive evidence base concerning small volume high cost groups such as ventilator-dependent children

The current studies found that the transfer of the ventilator from intensive care environments to new domestic settings for use by a different population group other than originally intended had resulted in unintended impacts and necessitated the need for a new and more extensive evidence base. For example:

- Existing expectations and clinical practices around the traditional use of the ventilator have been disturbed;
- The ventilator as a technology has evolved with new novel functionality resulting in the production of new modes and models, new markets and the need for new regulation;
- The new population of ventilator-dependent children have created their own novel 'technology-dependent' lifestyle which in turn has created

novel organisational demands, especially for resources and new services, staff require new skills and their knowledge needs to be redefined;

- The acceptance of children's dependence on machines to live has brought about the need for social and biological boundaries to be redefined, especially around parenting and technological care, new risks need to be conceptualised and managed, and
- Existing health and social policies concerning disabled children have been found to be inadequate when translated to children with such complex needs for health and social care; changes to policy are occurring long after long-term ventilation has become a routine intervention and transferred to domestic settings.

Although the Government has an established Health Technology Assessment programme to evaluate the effectiveness and cost-effectiveness of technologies, none of the evaluations of ventilators have looked in depth at the context of use or the unintended impacts of long-term use on children, their families, health services or society in general. One notable exception is the new Innovative Health Technology (IHT) programme that is funded jointly by the Economic and Social Research Council and Medical Research Council. This programme aims to commission research to determine the social consequences of medical technologies.

An additional IHT funded study that was carried out as part of the wider programme of research, but not reported in this thesis, looked at the implications concerning the use of established health technologies in new non-clinical settings (domestication of the ventilator and self-care, Heaton *et al.* 2003). Findings from Heaton *et al.* add to those reported in this thesis, that the scope of health technology assessments needs to be widened to determine how people and society will be affected by, and in turn affect, health technologies.

This thesis had a cross cutting methodological sub-theme to incorporate quantitative and qualitative methods into the overall research design and

synthesise findings across studies. The relevance of qualitative research to the assessment of health interventions has only recently been given recognition. At present, most systematic reviews of effectiveness are concerned with quantitative outcomes that can be meta-synthesised to calculate the magnitude of effect of a given intervention. The findings from qualitative research studies reported here have been used to help answer questions about the impact of long-term ventilation on health-related quality of life. These qualitative findings have the potential to enhance the scope, relevance and utility of quantitative systematic reviews concerning long-term ventilation and therapeutic interventions designed, for example, to improve health-related quality of life (Murphy *et al.* 1998). Further research is needed to refine integrative approaches to qualitative and quantitative evidence, but nonetheless there are now sufficiently rigorous methods for qualitative findings to be integrated into relevant quantitative systematic reviews of effectiveness to determine a wider assessment of efficacy and impacts of health technologies.

Funding of research into small volume high cost groups of children

The research programme reported in this thesis identified some important policy issues that arose from a very vulnerable small group of children and young people who were high consumers of health services. This research was expensive to carry out and almost all of the funding for fieldwork was derived from charities that had prioritised ventilator-dependent children as a small but socially excluded and under-researched group with many unmet needs for health and social care. There remains a danger that the specific needs of this and other small volume high cost groups of children and young people will be lost in the development of a population-based national health service for children. Specific funds need to be made available to undertake further primary research to add to the existing small evidence base, and secondary research to accumulate evidence in rigorously conducted mixed-method systematic reviews of effectiveness.

Likewise, the National Institute for Clinical Excellence (NICE) and the Social Care Institute for Excellence (SCIE) provide evidence based guidance on the

effectiveness and cost-effectiveness of interventions and services. Thus far, the majority of NICE recommendations around funding concern issues that are either contentious (such as in-vitro-fertilisation) or are of importance to public health generally (such as pharmaceutical products). NICE are unlikely to see the effectiveness and cost-effectiveness of long-term ventilation in children as a national priority. It would, however, be of great benefit for parents and doctors if an organisation with the authority of NICE or SCIE would provide morally defensible policy recommendations concerning the allocation of resources for ventilator-dependent children and their families.

10.3 Legal imperatives versus an evidence base

Meeting even the basic needs of children and young people dependent on ventilators costs money and all health and social care systems are constrained by finite resources (Glennister, 2003). Who pays, and who should pay is a matter of disagreement between the people involved, including:

- Parents who want resources for their children and believe that the state should provide them;
- Managers who have finite budgets and are required to balance the needs of small volume high cost groups such as ventilator-dependent children against the needs of entire populations;
- Government who manage the economy and have to determine health and social policy, and
- Taxpayers who do not want to be overburdened.

Parents were clearly concerned with the welfare of their children and focused on the need to maximise the wellbeing of their child. Parents defined subjectively the needs of their child and family, and described the resources and services that they would require in order to increase their autonomy and wellbeing. This 'welfarist' approach aligns with Doyal and Gough's (1991) theory of human need. They argue that all policies should start from the presumption that individuals (or families) ought to be able to survive and live

healthy autonomous lives. An important element in designing a social policy is that of 'agency', or the sense of being in control over one's own destiny (Glennister, 2003). This sense of control was evidently lacking in the lives of nearly all children and their families, and had a profound negative impact on their wellbeing. Parents clearly conceptualised the welfare of their child and wider family as a 'right' and some had used Human Rights legislation to increase the resources available to them, thereby increasing their sense of wellbeing and autonomy. As discussed previously in Chapter 9, direct payments to families may bring about the autonomy they desire if the money can be used to purchase a range of health and personal services of their choosing.

In contrast, doctors and managers focused more narrowly on 'health' and specifically on maximising the 'health' of the child as proxied with quantitative, utility-weighted measures. Doctors needed to assess whether a child had the ability to benefit from long-term ventilation as an intervention and yet there is no agreed definition of 'benefit' or an agreed way of measuring it.

This thesis highlights the tension between these two perspectives and the qualitative and quantitative evidence provides a different, often contrasting, picture concerning the health and wellbeing of the children and their families. In practice, there were many inequalities and inconsistencies in the level and range of services provided. Some children received packages of care that were designed to maximise their health and wellbeing, whereas other packages of care focused exclusively on maximising health gain from the ventilator.

The issue as to whether Government policy should aim to maximise welfare/wellbeing or more narrowly maximise health is not exclusive to ventilator-dependent children. Similar tensions exist with other patient groups. For example, in relation to the care of older people in England, current Government policy is to provide free health care to meet health needs, whereas the patient has to pay for, or contribute to, services to meet their personal care needs.

In relation to ventilator-dependent children, the unresolved dilemma remains as to what level of resources to bring about which desired outcomes should be provided by the state for children who either require ventilation as a short-term intervention towards the end of life, or for children who require ventilation as a long-term intervention over their lifespan.

The legal context to care and decision-making has evolved during the course of this programme of research. There have been major changes to the law concerning both children's rights to treatment (especially disabled children's rights), guidance on withdrawing and withholding treatment; and test cases that have deliberated on children's rights to healthcare and finite resources.

The conflicting claims around providing resources (or not) for ventilator-dependent children have transformed into an ethical problem as rationing or prioritisation of resources inevitably leads to policies that require difficult, even tragic, treatment decisions. Purtilo (1986) emphasises that as an arbiter amongst conflicting claims on resources, justice first requires that allocations are made to morally defensible criteria of distribution rather than on a random or arbitrary basis, or in situations whereby excessive pressure is exerted by parents.

As outlined in Chapter 6, the Court of Appeal has issued guidance on the general principles to be applied in establishing funding priorities (*R v North West Lancashire Health Authority*, 2000). The Appeal judges held that: ***'in establishing the respective needs of patients suffering from different illnesses and determining the respective strengths of their claims to treatment – it is vital for an Authority to:***

- ***Accurately assess the nature and seriousness of each type of illness;***
- ***Determine the effectiveness of various forms of treatment for it, and***
- ***Give proper effect to that assessment and that determination in the formulation and application of its policy.'***

This judgment reinforces the need to use an evidence base as an arbiter to determine an individual's rights to resources. Doctors in the current studies however did not have sufficient evidence concerning the magnitude of costs and benefits for all groups of children, they did not routinely refer difficult cases to the High Court, and they found it easier to comply with parents' wishes around active intervention.

In the absence of an adequate evidence base to inform decision-making, parents had used the judicial process to secure access to resources that had been denied them previously through other routes, irrespective of the ability of the NHS to pay or the potential benefit or disbenefit of long-term ventilation. Some children had also received compensation in response to complaints or as a result of the outcome of judicial reviews when children's rights were deemed to have been violated.

In the final days of completing this thesis, the European Court of Human Rights (ECHR) passed judgment on the case of a disabled child with severe sensory and physical impairments who required 24 hour care, that clarify some of the ethical issues raised by the findings of the current studies (*Glass v United Kingdom*, 2004).

In the ECHR case, doctors considered that the child in question was dying of respiratory failure and felt that instituting palliative and supportive care would be in the best interests of the child. The parents wanted all active measures to be taken to maintain their child's life including resuscitation and the initiation of ventilation. The doctors declined to resuscitate the child and the parents initiated legal action claiming that Under Article 8 of the Human Rights Act their child had the right to respect for his private and family life, his home and his correspondence. More specifically, Article 8 contends that:

'There shall be no interference by a public authority with the exercise of this right except such as is in accordance with the law and is necessary in a democratic society in the interests of national security, public safety or the economic wellbeing of the country, for the prevention of disorder or

crime, for the protection of health or morals, or for the protection of the rights and freedoms of others.'

In their deliberation the ECHR held that there had been a violation of Article 8 of the Convention and ordered that the child and his mother should be paid compensation by the UK Government. The judgement was based on the fact that the NHS Trust had not taken the case to the High Court for a legal opinion on the benefits or disbenefits of treating the child and what constituted 'the best interests of the child' in this case. This judgment reinforces the need for robust evidence around the costs and benefits of care that can be applied in individual cases, which must be referred to the High Court for deliberation.

In making their judgment, the judges reiterated that in such cases:

'... where there is reasonable uncertainty about the benefit of life-prolonging treatment, there should be a presumption in favour of initiating it, although there are circumstances in which active intervention (other than basic care) would not be appropriate since best interests is not synonymous with prolongation of life ... If the child's condition is incompatible with survival or where there is broad consensus that the condition is so severe that treatment would not provide a benefit in terms of being able to restore or maintain the patient's health, intervention may be unjustified. Similarly, where treatments would involve suffering or distress to the child, these and other burdens must be weighed against the anticipated benefit, even if life cannot be prolonged without treatment.'

The judgement reiterated that:

'The law has confirmed that best interests and the balance of benefits and burdens are essential components of decision-making and that the views of parents are a part of this. However, parents cannot necessarily insist on enforcing decisions based solely on their own preferences where these conflict with good medical evidence.'

This judgment asserted that doctors must take cases to the High Court for clarification, which will inevitably have considerable cost implications for the NHS. Evidence from the current studies however found that doctors regarded the legal route to withholding and withdrawing treatment as very distressing for both doctors and parents; doctors tended to adopt a neutral position with a 'wait and see' approach and go along with parent's wishes even though they may not be in the best interests of the child and not a prudent use of scarce resources.

Glass v United Kingdom (2004) also illustrates just how stressful events can be for the family and doctors concerned when a decision about treatment is contested and the case has not yet been decided by the High Court. In this particular case, despite prolonged discussions, the mother did not accept that her child should not be resuscitated and placed on a ventilator. When her child rapidly deteriorated and appeared to be in terminal respiratory failure and dying, she tried to resuscitate him whilst a fight broke out between members of the family and the doctors in attendance.

In Canada and the United States doctors and patients are supported in their decision-making by clinical ethicists (Sokol, 2003). Whereas disability rights campaigners such as Campbell (who is Chair of the Institute for Social Care Excellence in the UK and a ventilator user herself), have called for dedicated nurses for disabled people to challenge treatment decisions that do not favour intervention (Prasad, 2003). Campbell was reported as saying that such drastic measures are required as her doctors assumed that once she fell into a coma due to respiratory failure that she would no longer want treatment, when in fact she wanted full and active treatment whatever the consequences. Whatever the model of advocacy adopted, evidence from the current studies indicated that doctors, patients and parents alike appear to need more support than currently available to manage these extremely difficult and sensitive situations.

Interestingly, in their deliberation, the ECHR judges did not consider that **'the economic wellbeing of the country'** constituted an exception to not upholding Article 8 in this case. Nor did they provide any guidance on what constituted **'good medical evidence'**. Indeed, in England, parents have previously gained

resources to purchase packages of home care for their children based on the same Article (8), as their right to a family life had been violated by the continued hospitalisation of their children (irrespective of the benefits or disbenefits of the intervention) (for example, *N v Department of Health In Northern Ireland*, 2003).

As highlighted at the beginning of this chapter, there remains an urgent need for a more extensive evidence base especially around the magnitude of benefit conferred by long-term ventilation with different groups of children, the changes to health-related quality of life over time, outcomes of health and social care, and the most effective ways to deliver services. This programme of research, however, goes some way to providing new evidence around the costs and social benefits of caring for children dependent on ventilators.

Appendix 1 - The KINDL and EQ5D Instruments

ID:

Questionnaire for children

Age 6 - 7 years

Hello there!

We would like to know how you have been feeling, so we have put together a few questions that we would like you to answer.

- ➔ I am going to read each question to you
- ➔ Think about how things have been for you
- ➔ Tell me which answer fits you best

There are no right or wrong answers. It's what you think that matters.

Today's Date: ___ / ___ / ___ (day / month / year)

Section 1. First of all, please tell me something about you

Are you a..... girl boy

How old are you? years

How many brothers and sisters do you have? 0 1 2 3
4 5 more than 5

Which school do you attend?

Which year are you in at school? Year

Where are you currently living? Family home Hospital
Other (please state where).....

Section 2. Thinking about the past WEEK

Now, I will read you an example:

Listen to the following question:

How often during the past week did you feel like eating ice-cream?

Did you:

- NEVER felt like eating ice cream
- SOMETIMES felt like eating ice cream
- VERY OFTEN felt like eating ice cream

You did that very well. So now let us begin....

1. First of all, we would like to know something about your health...

During the past week....	Never	Sometimes	Very often
1. ...I felt ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I had a headache or tummy-ache	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

2. Then something about how you've been feeling in general....

During the past week....	Never	Sometimes	Very often
1. ... I had fun and laughed a lot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was bored	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

3.... And how are you feeling about yourself?

During the past week....	Never	Sometimes	Very often
1. ... I was proud of myself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I felt pleased with myself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

4.... The next questions are about your family life...

During the past week....	Never	Sometimes	Very often
1. ... I got on well with my parent(s)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was happy where I was living	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

5. ... And then about friends...

During the past week....	Never	Sometimes	Very often
1. ... I played with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I got along well with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

6. ... We would like to know something about school ...

During the past week....	Never	Sometimes	Very often
1. ... I found my work at school was easy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I enjoyed nursery school/school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7. ... We would like to know something about you...

During the past week....	Never	Sometimes	Very often
1. ... I was afraid that my health might get worse	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was sad because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I was able to cope well with my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My parent(s) treated me like a baby because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. I missed something at nursery school/school because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. I avoided others because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Section 3. Describing your health TODAY

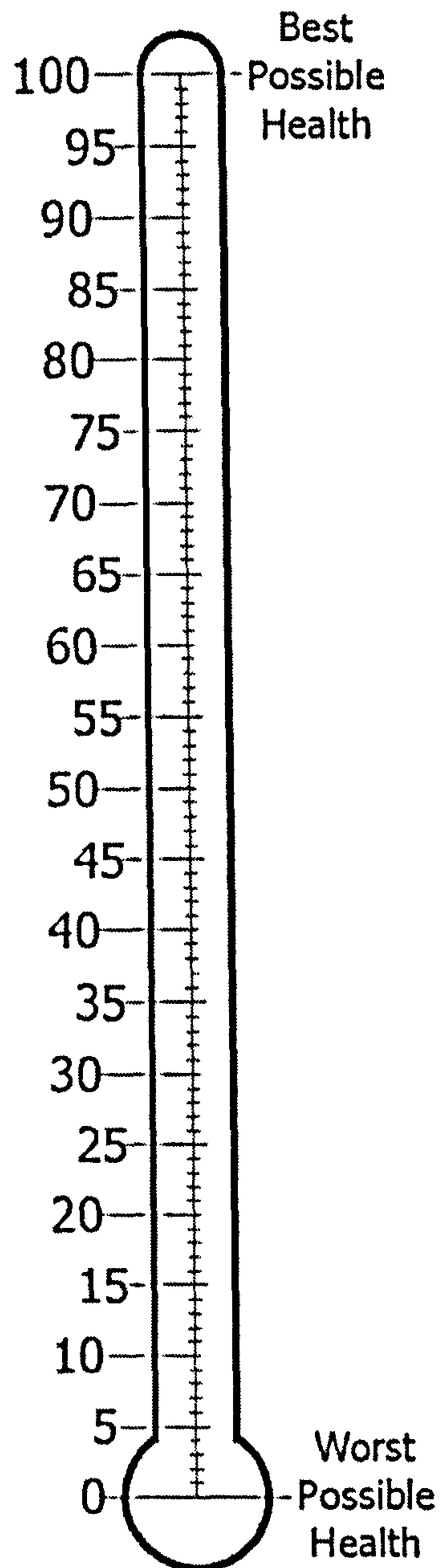
Which of the following statements fits you best?

My mobility <u>TODAY</u>	
I have no problems walking about	<input type="checkbox"/>
I have some problems walking about	<input type="checkbox"/>
I have a lot of problems walking about	<input type="checkbox"/>
I cannot walk about	<input type="checkbox"/>
Looking after myself <u>TODAY</u>	
I have no problems washing or dressing myself	<input type="checkbox"/>
I have some problems washing or dressing myself	<input type="checkbox"/>
I am unable to wash or dress myself	<input type="checkbox"/>
My usual activities <u>TODAY</u> (eg Going to school, hobbies, sports, playing)	
I have no problems doing my usual activities	<input type="checkbox"/>
I have some problems doing my usual activities	<input type="checkbox"/>
I am unable to do my usual activities	<input type="checkbox"/>
Pain/Discomfort <u>TODAY</u>	
I have no pain or discomfort	<input type="checkbox"/>
I have some pain or discomfort	<input type="checkbox"/>
I have a lot of pain or discomfort	<input type="checkbox"/>
Feeling worried, sad or unhappy <u>TODAY</u>	
I am not worried, sad or unhappy	<input type="checkbox"/>
I am a bit worried, sad or unhappy	<input type="checkbox"/>
I am very worried, sad or unhappy	<input type="checkbox"/>

Section 4. How good is your health TODAY?

Please draw a line through the scale at the point which best describes your health TODAY.

- The best possible health you can imagine is marked 100
- The worst possible health you can imagine is marked 0

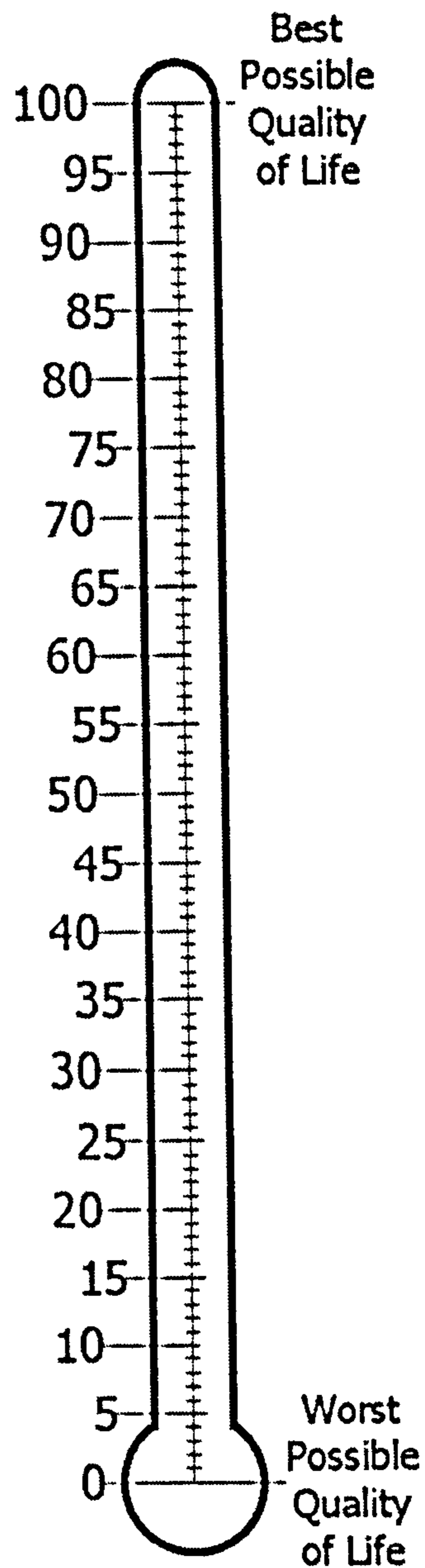


Section 5: How good is your quality of life TODAY?

Now we would like to ask about your overall quality of life.

Please draw a line through the scale at the point which best describes your quality of life TODAY.

- The best possible quality of life you can imagine is marked 100
- The worst possible quality of life you can imagine is marked 0



Section 6: How good is your health TODAY compared to other children of your age?

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 7: How good is your quality of life TODAY compared to other children of your age?

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 8: Can you name some things that affect your quality of life?

Good things	Bad things
1.	1.
2.	2.
3.	3.
4.	4.

Thank you for helping us!

ID:

Questionnaire for children

Age 8 and over

Hello there!

We would like to know how you have been feeling, so we have put together a few questions that we would like you to answer.

- ➔ Please read each question carefully.
- ➔ There are no right or wrong answers. It's what you think that matters.

Section 1. About Yourself

(please put a cross in the appropriate box or fill in details)

Are you a..... girl boy

How old are you? years

How many brothers and sisters do you have? 0 1 2 3
4 5 more than 5

Which school do you attend?

Which year are you in at school? Year

Where are you currently living? Family home Hospital
Other (please state where).....

Today's Date: ___ / ___ / ___ (day / month / year)

Section 2. Thinking about the PAST WEEK

➔ Please put a cross in the box corresponding to the answer that fits you best.

<u>For example:</u>	Never	Rarely	Some- times	Often	All the time
During the <u>PAST WEEK</u> , I felt like eating ice cream	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input checked="" type="checkbox"/>

1. First of all, we would like to know something about your physical health...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I felt ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was in pain	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I was tired and worn out	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I felt strong and full of energy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

2. Then something about how you've been feeling in general...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I had fun and laughed a lot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was bored	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I felt alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I felt scared	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

3... And how are you feeling about yourself?

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I felt proud of myself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I felt on top of the world	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I felt pleased with myself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I had lots of good ideas	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

4. ... The next questions are about your family ...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I got on well with my parent(s)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I felt fine where I was living	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I quarrelled with my family	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My parent(s) stopped me from doing certain things	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

5. ... And then about friends...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I played with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was liked by my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I got along well with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I felt different from other children and young people	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

6. ... We would like to know something about school ...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... I found doing my work at school was easy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I found school interesting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I worried about my future	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... I worried about getting bad marks or grades at school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7. ... We would like to know something about you...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... I was afraid that my health might get worse	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was sad because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I was able to cope well with my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4 ... My parent(s) treated me like a baby because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5 ... I avoided others because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6 ... I missed something at school because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Section 3: Describing your health TODAY

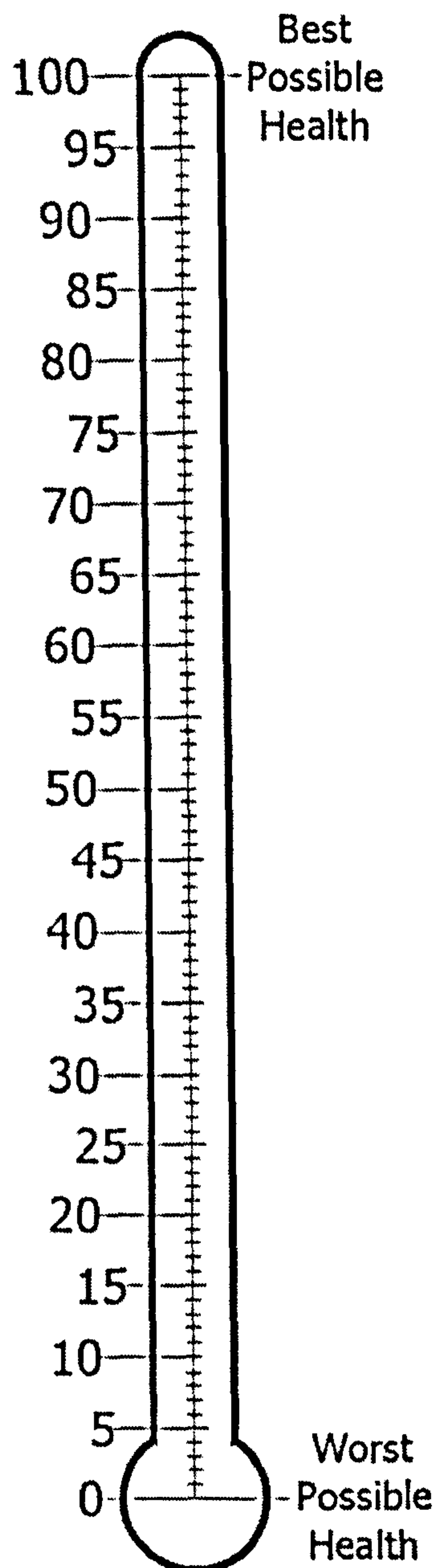
- Please put a cross in ONE box in each section that best describes your health TODAY

My mobility <u>TODAY</u>	
I have no problems walking about	<input type="checkbox"/>
I have some problems walking about	<input type="checkbox"/>
I have a lot of problems walking about	<input type="checkbox"/>
I cannot walk about	<input type="checkbox"/>
Looking after myself <u>TODAY</u>	
I have no problems washing or dressing myself	<input type="checkbox"/>
I have some problems washing or dressing myself	<input type="checkbox"/>
I am unable to wash or dress myself	<input type="checkbox"/>
My usual activities <u>TODAY</u> (eg Going to school, hobbies, sports, playing)	
I have no problems doing my usual activities	<input type="checkbox"/>
I have some problems doing my usual activities	<input type="checkbox"/>
I am unable to do my usual activities	<input type="checkbox"/>
Pain/Discomfort <u>TODAY</u>	
I have no pain or discomfort	<input type="checkbox"/>
I have some pain or discomfort	<input type="checkbox"/>
I have a lot of pain or discomfort	<input type="checkbox"/>
Feeling worried, sad or unhappy <u>TODAY</u>	
I am not worried, sad or unhappy	<input type="checkbox"/>
I am a bit worried, sad or unhappy	<input type="checkbox"/>
I am very worried, sad or unhappy	<input type="checkbox"/>

Section 4: How good is your health TODAY?

Please draw a line through the scale at the point which best describes your health TODAY.

- The best possible health you can imagine is marked 100
- The worst possible health you can imagine is marked 0



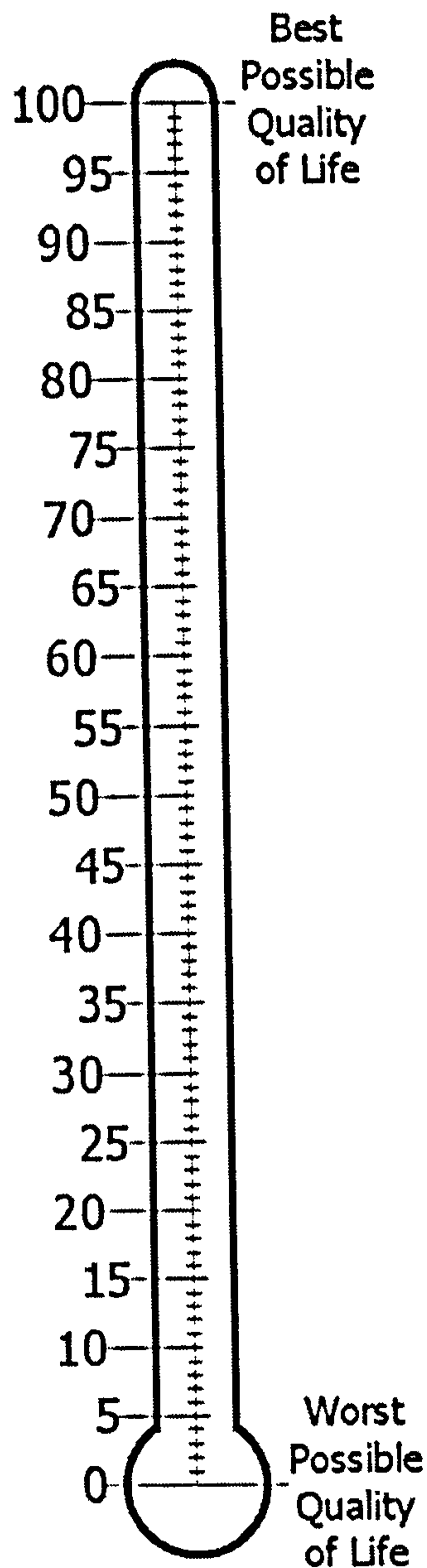
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Section 5. How good is your quality of life TODAY?

Now we would like to ask about your overall quality of life.

Please draw a line through the scale at the point which best describes your quality of life TODAY.

- The best possible quality of life you can imagine is marked 100
- The worst possible quality of life you can imagine is marked 0



Section 6. How good is your health TODAY compared to other children of your age?
 (Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 7. How good is your quality of life TODAY compared to other children of your age?
 (Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 8. Can you name some things that affect your quality of life? (Please write comments)

Good things	Bad things
1.	1.
2.	2.
3.	3.
4.	4.

Thank you for helping us!

ID:

Questionnaire for young people

Age 13 years +

Hello there!

We would like to know how you have been feeling, so we have put together a few questions that we would like you to answer.

- ➔ Please read each question carefully
- ➔ There are no right or wrong answers. It's what you think that matters.

Section 1. About Yourself

(please put a cross in the appropriate box or fill in details)

Are you a..... girl boy

How old are you? years

How many brothers and sisters do you have? 0 1 2 3
4 5 more than 5

Which school do you attend?

Which year are you in at school? Year

Where are you currently living? Family home Hospital

Other (please state where).....

Today's Date: ___ / ___ / ___ (day / month / year)

Section 2. Thinking about the PAST WEEK

➔ Please put a cross in the box corresponding to the answer that fits you best.

For example:	Never	Rarely	Some- times	Often	All the time
During the <u>PAST WEEK</u> , I felt like eating ice cream	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input checked="" type="checkbox"/>

1. First of all, we would like to know something about your physical health...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I felt ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was in pain	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I was tired and worn out	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I felt strong and full of energy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

2. Then something about how you've been feeling in general....

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I had fun and laughed a lot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was bored	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I felt alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I felt scared or unsure of myself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

3. ... And how are you feeling about yourself?

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I felt proud about the things I did	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I felt on top of the world	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I felt pleased with myself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I had lots of good ideas	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

4. ... The next questions are about your family ...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I got on well with my parent(s)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I felt fine where I was living	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I quarrelled with my family	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My parent(s) stopped me from doing certain things	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

5. ... And then about friends...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ...I did things together with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was popular with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I got along well with my friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ...I felt different from other children and young people	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

6. ... We would like to know something about school ...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... I found doing my schoolwork was easy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I found school interesting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I worried about my future	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... I worried about getting bad marks or grades at school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7. ... We would like to know something about you...

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... I was afraid that my health might get worse	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... I was sad because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I was able to cope well with my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4 ... My parent(s) treated me like a baby because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5 ... I avoided others because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6 ... I missed something at school because of my health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Section 3: Describing your health TODAY

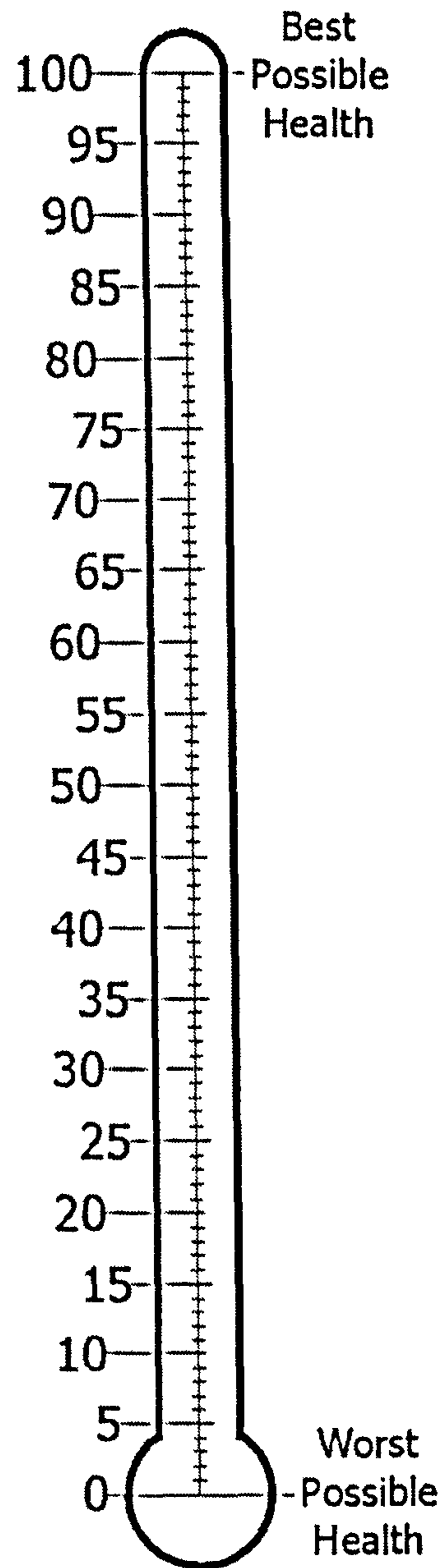
➔ Please put a cross in ONE box in each section that best describes your health TODAY

My mobility <u>TODAY</u>	
I have no problems walking about	<input type="checkbox"/>
I have some problems walking about	<input type="checkbox"/>
I have a lot of problems walking about	<input type="checkbox"/>
I cannot walk about	<input type="checkbox"/>
Looking after myself <u>TODAY</u>	
I have no problems washing or dressing myself	<input type="checkbox"/>
I have some problems washing or dressing myself	<input type="checkbox"/>
I am unable to wash or dress myself	<input type="checkbox"/>
My usual activities <u>TODAY</u> (eg Going to school, hobbies, sports, playing)	
I have no problems doing my usual activities	<input type="checkbox"/>
I have some problems doing my usual activities	<input type="checkbox"/>
I am unable to do my usual activities	<input type="checkbox"/>
Pain/Discomfort <u>TODAY</u>	
I have no pain or discomfort	<input type="checkbox"/>
I have some pain or discomfort	<input type="checkbox"/>
I have a lot of pain or discomfort	<input type="checkbox"/>
Feeling worried, sad or unhappy <u>TODAY</u>	
I am not worried, sad or unhappy	<input type="checkbox"/>
I am a bit worried, sad or unhappy	<input type="checkbox"/>
I am very worried, sad or unhappy	<input type="checkbox"/>

Section 4: How good is your health TODAY?

Please draw a line through the scale at the point which best describes your health TODAY.

- The best possible health you can imagine is marked 100
- The worst possible health you can imagine is marked 0

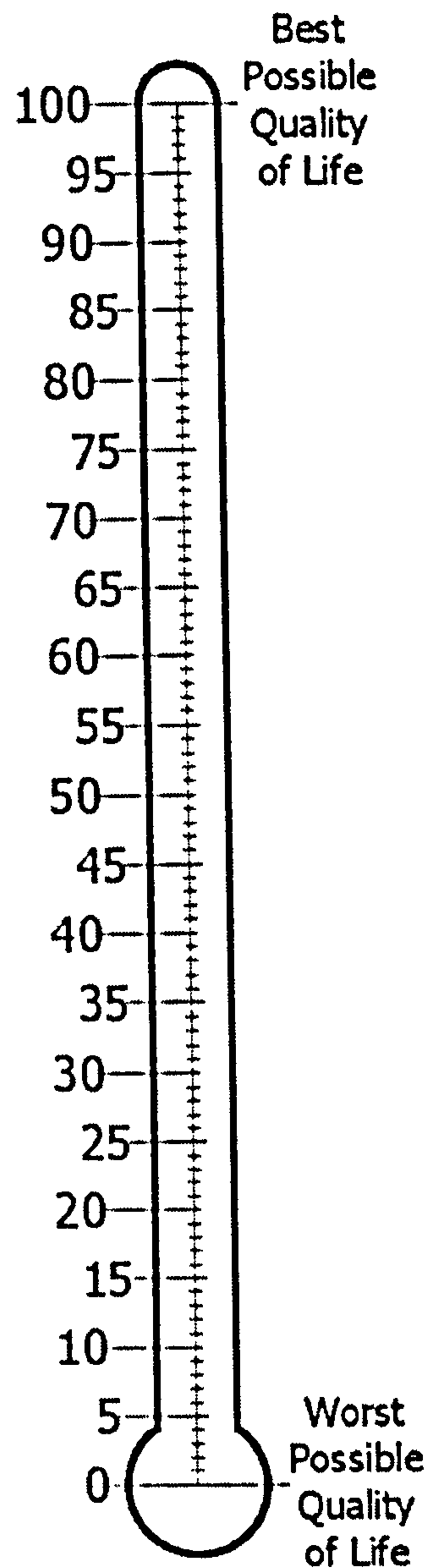


**Section 5. How good is your quality of life
TODAY?**

Now we would like to ask about your overall quality of life.

Please draw a line through the scale at the point which best describes your quality of life TODAY.

- The best possible quality of life you can imagine is marked 100
- The worst possible quality of life you can imagine is marked 0



Section 6. How good is your health TODAY compared to other children of your age?

(Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 7. How good is your quality of life TODAY compared to other children of your age?

(Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 8. Can you name some things that affect your quality of life? (Please write comments)

Good things	Bad things
1.	1.
2.	2.
3.	3.
4.	4.

Thank you for helping us!

ID:

Questionnaire for parents of children aged 4-7 years

Dear Parent,

Thank you for taking the time to complete this questionnaire about your child's well-being and health-related quality of life.

Since it is a matter of your own assessment of your child's well being, please complete the questionnaire yourself according to the instructions and without asking your child.

- ➔ Please read each question carefully.
- ➔ There are no right or wrong answers. It's what you think that matters.

Section 1. About you and your child

(please put a cross in the appropriate box or fill in details)

My child is a: Girl Boy Age:.....years

I am the child's: Mother Father Other.....

How many brothers and sisters does your child have? 0 1 2
3 4 5+

Today's Date: ___ / ___ / ___ (day / month / year)

Section 2. Think about how your child has been feeling during the PAST WEEK.

➔ Please put a cross in the box corresponding to the answer that best fits your child.

<u>For example:</u>	Never	Rarely	Some-times	Often	All the time
During the <u>PAST WEEK</u> , my child felt like eating ice cream	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input checked="" type="checkbox"/>

1. Physical Well-being

During the <u>PAST WEEK</u>	Never	Rarely	Some-times	Often	All the time
1. ... My child felt ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child had a headache or tummy-ache	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child was tired and worn out	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt strong and full of energy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

2. Emotional Well-being

During the <u>PAST WEEK</u>	Never	Rarely	Some-times	Often	All the time
1. ... My child had fun and laughed a lot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child didn't feel much like doing anything	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child felt alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt scared or unsure of him/her self	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

3. Self Esteem

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child was proud of him/her self	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child felt on top of the world	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child felt pleased with him/her self	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child had lots of good ideas	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

4. Family

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child got on well with us as parent(s)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child felt fine where he/she was living	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I quarrelled with my child	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt that I was bossing him/her around	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

5. Social contacts

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child played with his/her friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child was liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child got along well with his/her friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt different from other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

6. School

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child coped easily with activities at nursery school/school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child enjoyed nursery school/school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child looked forward to nursery school/school	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child made lots of mistakes when doing nursery/school activities	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7. Health

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child was afraid that his/her health might get worse	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child was sad because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child was able to cope well with his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4 ... I treated my child as though he/she was younger because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5 ... My child avoided others because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6 ... My child missed something at school because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

8. Additional questions

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child was moody and whined a lot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child had a healthy appetite	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I managed to show patience and understanding towards my child	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4 ... My child felt under pressure	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5 ... My child slept soundly	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6 ... My child was very active	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. ... My child kept bursting into tears	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. ... My child was cheerful and in a good mood	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. ... My child was alert and able to concentrate well	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10. ... My child was easily distracted and absent minded	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
11... My child enjoyed being with other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
12. I had to give my child a telling off	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
13. I praised my child	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
14. My child had problems with teachers, carers or other child minders	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
15 ... My child was nervous and fidgety	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
16. ... My child was lively and energetic	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
17. ... My child was sociable and outgoing	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
18. ... My child succeeded at everything he set out to do	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
19... My child became dissatisfied easily	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
20... My child cried bitterly	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
21... My child lost his/her temper quickly	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Section 3: Describing your child's health TODAY

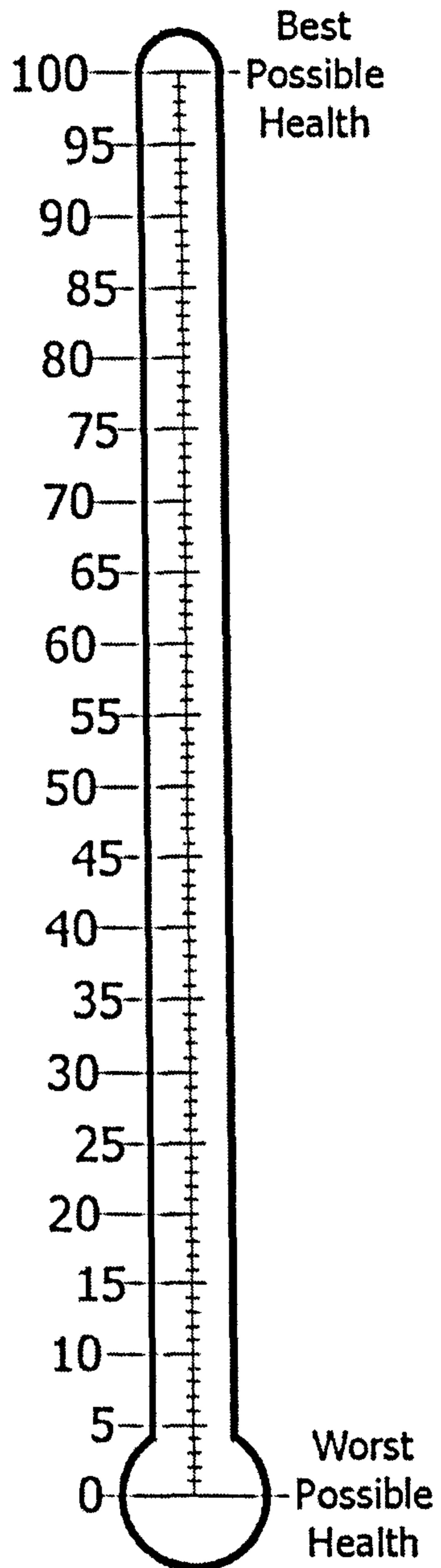
- ➔ Please put a cross in ONE box in each section that best describes your child's health TODAY.

Mobility	
He/she has no problems walking about	<input type="checkbox"/>
He/she has some problems walking about	<input type="checkbox"/>
He/she has a lot of problems walking about	<input type="checkbox"/>
He/she cannot walk about	<input type="checkbox"/>
Looking after himself or herself	
He/she has no problems washing or dressing him/herself	<input type="checkbox"/>
He/she has some problems washing or dressing him/herself	<input type="checkbox"/>
He/she is unable to wash or dress him/herself	<input type="checkbox"/>
Usual activities (eg Going to school, hobbies, sports, playing)	
He/she has no problems doing his/her usual activities	<input type="checkbox"/>
He/she has some problems doing his/her usual activities	<input type="checkbox"/>
He/she is unable to do his/her usual activities	<input type="checkbox"/>
Pain/Discomfort	
He/she has no pain or discomfort	<input type="checkbox"/>
He/she has some pain or discomfort	<input type="checkbox"/>
He/she has a lot of pain or discomfort	<input type="checkbox"/>
Feeling worried, sad or unhappy	
He/she is not worried, sad or unhappy	<input type="checkbox"/>
He/she is a bit worried, sad or unhappy	<input type="checkbox"/>
He/she is very worried, sad or unhappy	<input type="checkbox"/>

Section 4. How good is your child's health TODAY?

Please draw a line through the scale at the point which best describes your child's health TODAY.

- The best possible health you can imagine is marked 100
- The worst possible health you can imagine is marked 0

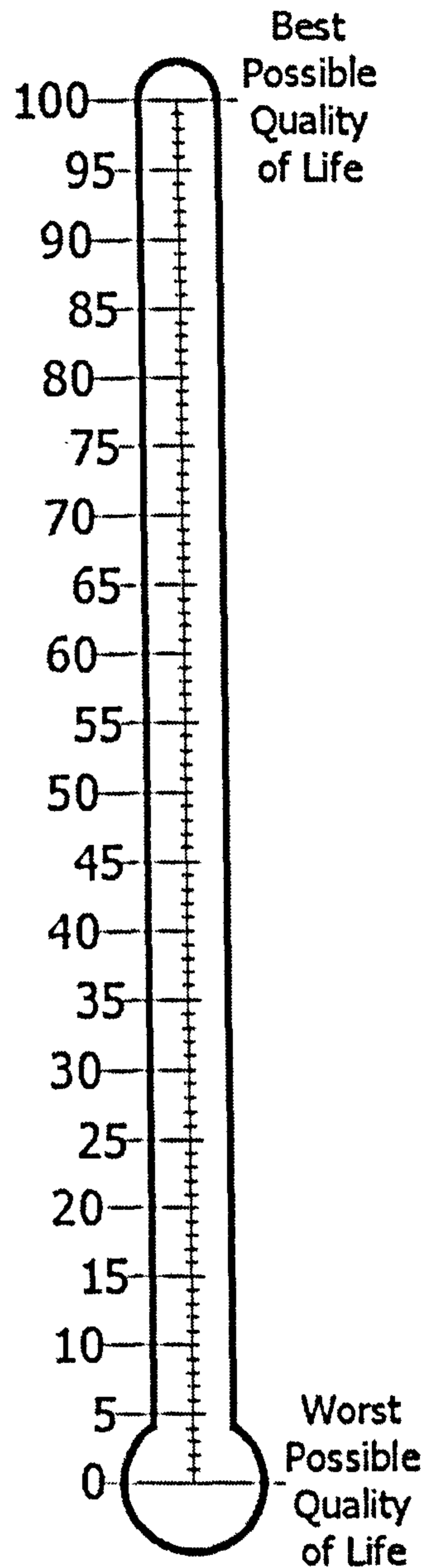


Section 5. How good is your child's quality of life TODAY?

Now we would like to ask about your child's overall quality of life.

Please draw a line through the scale at the point which best describes your child's quality of life TODAY.

- The best possible quality of life you can imagine is marked 100
- The worst possible quality of life you can imagine is marked 0



Section 6. How good is your child's health TODAY compared to other children of his/her age?
 (Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 7. How good is your child's quality of life TODAY compared to other children of his/her age?
 (Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 8. Can you name some things that affect your child's quality of life? (Please write comments)

Good things	Bad things
1.	1.
2.	2.
3.	3.
4.	4.

Thank you for helping us!

ID:

Questionnaire for parents of children aged 8 years and over

Dear Parent,

Thank you for taking the time to complete this questionnaire about your child's well-being and health-related quality of life.

Since it is a matter of your own assessment of your child's well being, please complete the questionnaire yourself according to the instructions and without asking your child.

- ➔ Please read each question carefully.
- ➔ There are no right or wrong answers. It's what you think that matters.

Section 1. About you and your child

(please put a cross in the appropriate box or fill in details)

My child is a: Girl Boy Age:.....years

I am the child's: Mother Father Other.....

How many brothers and sisters does your child have? 0 1 2
3 4 5+

Today's Date: ___ / ___ / ___ (day / month / year)

Section 2. Think about how your child has been feeling during the PAST WEEK.

➔ Please put a cross in the box corresponding to the answer that best fits your child.

<u>For example:</u>	Never	Rarely	Some-times	Often	All the time
During the <u>PAST WEEK</u> , my child felt like eating ice cream	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input checked="" type="checkbox"/>

1. Physical Well-being

During the <u>PAST WEEK</u>	Never	Rarely	Some-times	Often	All the time
1. ... My child felt ill	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child was in pain	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child was tired and worn out	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt strong and full of energy	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

2. Emotional Well-being

During the <u>PAST WEEK</u>	Never	Rarely	Some-times	Often	All the time
1. ... My child had fun and laughed a lot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child didn't feel much like doing anything	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child felt alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt scared or unsure of him/her self	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

3. Self Esteem

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child was proud of him/her self	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child felt on top of the world	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child felt pleased with him/her self	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child had lots of good ideas	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

4. Family

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child got on well with us as parent(s)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child felt fine where he/she was living	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... I quarrelled with my child	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt that I was bossing him/her around	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

5. Social contacts

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child played with his/her friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child was liked by other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child got along well with his/her friends	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child felt different from other children	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

6. School

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child coped easily with schoolwork	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child found lessons at school interesting	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child worried about his/her future	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... My child was worried about getting bad marks or grades	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

7. Health

During the <u>PAST WEEK</u>	Never	Rarely	Some- times	Often	All the time
1. ... My child was afraid that his/her health might get worse	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. ... My child was sad because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. ... My child was able to cope well with his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. ... I treated my child as though he/she was younger because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. ... My child avoided others because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. ... My child missed something at school because of his/her health	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Section 3: Describing your child's health TODAY

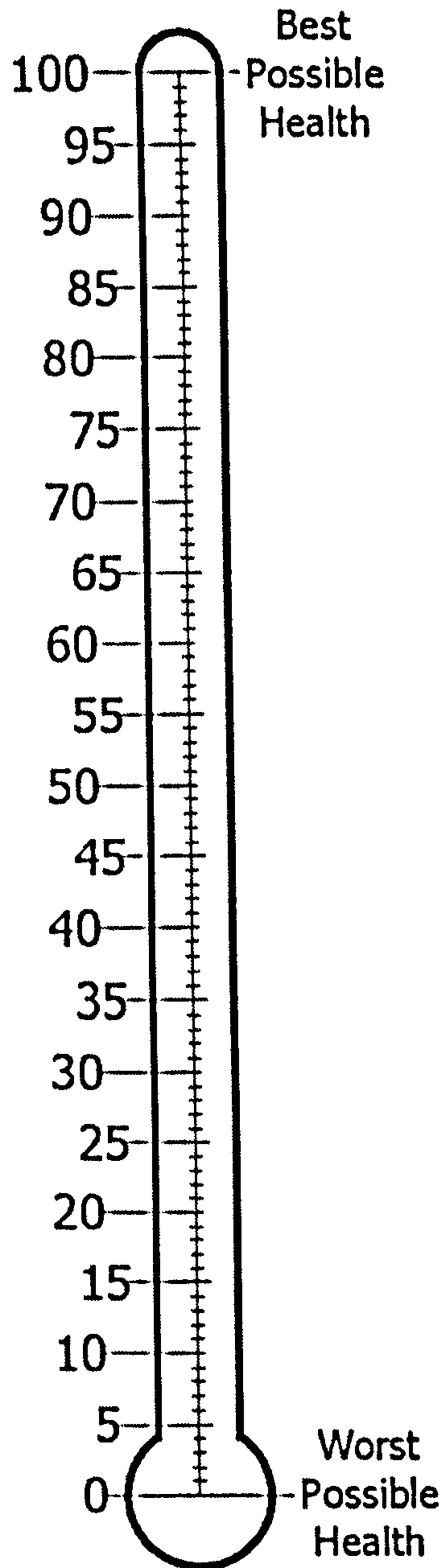
➔ Please put a cross in ONE box in each section that best describes your child's health TODAY

Mobility	
He/she has no problems walking about	<input type="checkbox"/>
He/she has some problems walking about	<input type="checkbox"/>
He/she has a lot of problems walking about	<input type="checkbox"/>
He/she cannot walk about	<input type="checkbox"/>
Looking after himself or herself	
He/she has no problems washing or dressing him/herself	<input type="checkbox"/>
He/she has some problems washing or dressing him/herself	<input type="checkbox"/>
He/she is unable to wash or dress him/herself	<input type="checkbox"/>
Usual activities (eg Going to school, hobbies, sports, playing)	
He/she has no problems doing his/her usual activities	<input type="checkbox"/>
He/she has some problems doing his/her usual activities	<input type="checkbox"/>
He/she is unable to do his/her usual activities	<input type="checkbox"/>
Pain/Discomfort	
He/she has no pain or discomfort	<input type="checkbox"/>
He/she has some pain or discomfort	<input type="checkbox"/>
He/she has a lot of pain or discomfort	<input type="checkbox"/>
Feeling worried, sad or unhappy	
He/she is not worried, sad or unhappy	<input type="checkbox"/>
He/she is a bit worried, sad or unhappy	<input type="checkbox"/>
He/she is very worried, sad or unhappy	<input type="checkbox"/>

Section 4. How good is your child's health TODAY?

Please draw a line through the scale at the point which best describes your child's health TODAY.

- The best possible health you can imagine is marked 100
- The worst possible health you can imagine is marked 0

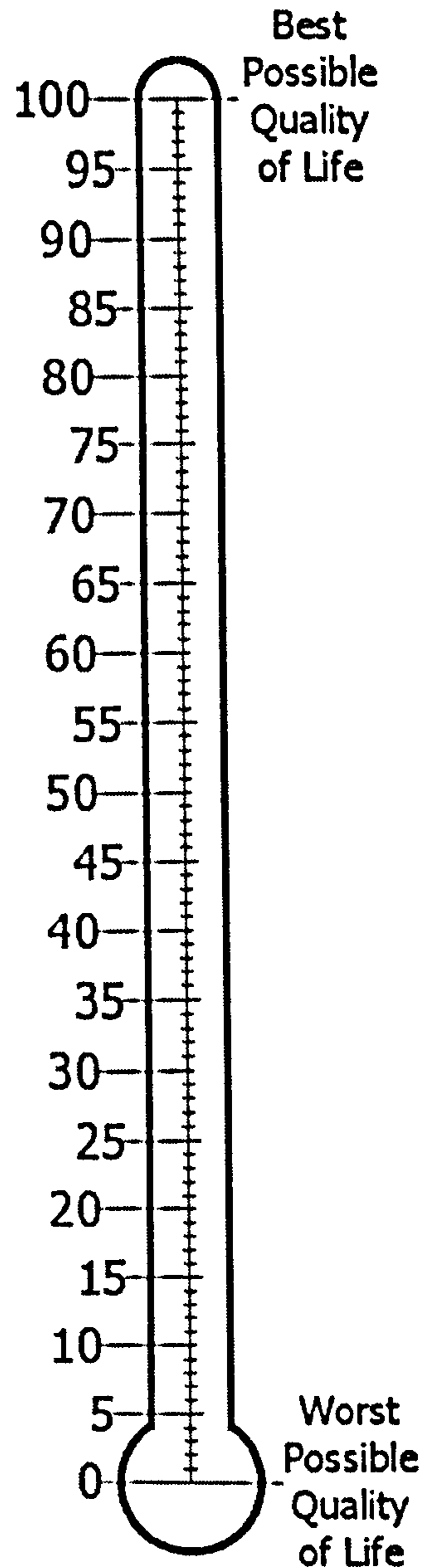


Section 5. How good is your child's quality of life TODAY?

Now we would like to ask about your child's overall quality of life.

Please draw a line through the scale at the point which best describes your child's quality of life TODAY.

- The best possible quality of life you can imagine is marked 100
- The worst possible quality of life you can imagine is marked 0



Section 6. How good is your child's health TODAY compared to other children of his/her age?
 (Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 7. How good is your child's quality of life TODAY compared to other children of his/her age?
 (Please put a cross in the appropriate box)

1. Much better	<input type="checkbox"/>
2. Better	<input type="checkbox"/>
3. Same	<input type="checkbox"/>
4. Worse	<input type="checkbox"/>
5. Much worse	<input type="checkbox"/>

Section 8. Can you name some things that affect your child's quality of life? (Please write comments)

Good things	Bad things
1.	1.
2.	2.
3.	3.
4.	4.

Thank you for helping us!

Appendix 2 - Qualitative Findings Framed Around the Articles of the Human Rights Act 1998

The United Nations convention on the rights of the child

The main findings from the qualitative study exploring the experiences of ventilator-dependent children and their families (Chapters 4 and 7) were subsequently framed around the relevant articles of the UN Convention on the Rights of the Child (1989).

General measures of implementation:

The state has the duty to take all possible measures to ensure implementation of the Convention (Article 4) and the duty to make the principles of the Convention widely known to parents and children (Article 42). This study found that:

- Health, social and education departments had not fully implemented the Convention, and
- The young people and their parents had not been made fully aware of their rights.

Disabled children:

The state has a responsibility to ensure that disabled children enjoy a full and decent life in conditions which ensure dignity, promote self-reliance, and facilitate the child's active participation in the community (Article 23).

The study found that:

- Some young people lived inappropriately in hospitals, and
- Many young people were socially excluded.

Basic health and welfare:

The state has a responsibility to ensure the development of institutions, facilities and services for the care of children (Articles 18, 23). The child has the right to the highest attainable standard of health and to facilities for the treatment of illness and rehabilitation of health (Articles 23, 24). All parties must ensure the optimal development of each child (Articles 6, 23) and the state has an

obligation to protect children from any form of discrimination (Articles 2, 23). All actions concerning the child must take full account of his or her best interests (Article 3). This study found strong evidence that:

- Gross inequalities existed in the health and social care, provided by different regions;
- The young people were discriminated against because of their dependency on a ventilator and they found it difficult to access appropriate services;
- The young people were transferred to a number of NHS hospitals for treatment and rarely had continuity of care;
- Not all hospitals provided appropriate play, recreational activities and a programme of rehabilitation;
- The NHS, social services and education authorities were generally inefficient and inept at discharging these young people into the community;
- Care packages enabling the young people to live at home varied widely and were not based on need;
- The NHS, social services and education authorities demonstrated an inability to work together in the best interests of the young person;
- Some young people received incompetent care and treatment;
- Little transition planning had been undertaken by the NHS, social services and education authorities. This is not in the best interests of the child, and
- There was gross inefficiency and wastage of resources.

Social security:

Children have the right to benefit from social security. The benefits should be granted taking into account the unique circumstances of each child (Article 26). Every child has a right to an adequate standard of living and housing (Article 27). This study found that:

- Some young people were denied benefits because the application process did not take account of their individual unique circumstances;
- Some young people lived at home in unsuitable and unsafe housing, and
- Some young people were being accommodated on a long-term basis in healthcare settings, which were unsuitable and not inspected by social services departments.

Civil rights and freedoms:

The child has a right to express his or her own opinions and views (Article 12, 13) and access to appropriate information (Article 17). This includes freedom of thought, conscience, religion and association (Articles 14, 15). The child has the right to be protected from all forms of mal-treatment and other cruel inhuman or degrading treatment (Articles 19, 37a). This study found strong evidence that:

- Some young people did not have access to a communication system that suited their needs and they could not freely express their views;
- Some young people did not always have contact with people that understood the ways they communicated;
- The young people and their parents did not always have access to appropriate information and were frequently excluded from making important decisions about their lives;
- Most of the young people spent prolonged periods of time (sometimes years) in hospital when they no longer needed, or wanted, to be there. This deprived them of their freedom and contact with those they loved. This was considered to be cruel and inhumane;
- Some of the young people did not have parents or guardians to help make important decisions and were not appointed an Independent Visitor or an advocate independent of the system (as outlined in the Children Act), and
- The young people were frequently not respected, listened to, or acknowledged as experts.

The family:

The state has a duty to respect the rights and responsibilities of parents and the wider family (Articles 5, 7, 18). The child has a right to live with and maintain contact with both his/her parents (unless there are exceptional circumstances) and lead a full and active life in society (Articles 9, 23). The state has a responsibility to protect and if necessary re-establish the basic aspects of a child's identity (Articles 7, 8, 20, 30). The child and their family have a right to privacy (Article 16) and the state must periodically review their care and treatment (Article 25). This study found strong evidence that:

- Many professionals did not respect the rights and continued responsibilities of parents regarding their children;
- Not all parents were offered on site hospital accommodation to stay with their children (as outlined in the NHS Children's Charter);
- Not all professionals actively promoted the child's right to live with their family;
- Most young people, at some stage, were denied their right to live with their families because of unnecessarily prolonged hospitalisation;
- The young people were not always consulted regarding their need for continued contact with their parents and siblings;
- Some young people were uncertain of their life histories and biographies and did not have essential memories maintained for them;
- Some young people had lost contact with their language, religion, culture and roots;
- The young people in residential accommodation felt they did not belong to their families any more;
- The young people and their parents said that their right to privacy was frequently violated;
- Social services review meetings were infrequent and in some cases did not happen;
- On occasions the young people were reduced to object status, especially in meetings;

- Facilities for, and provision of, appropriate 'respite care' were totally inadequate, and
- Parents found that complaints procedures were inadequate and resorted to using the media and their Member of Parliament.

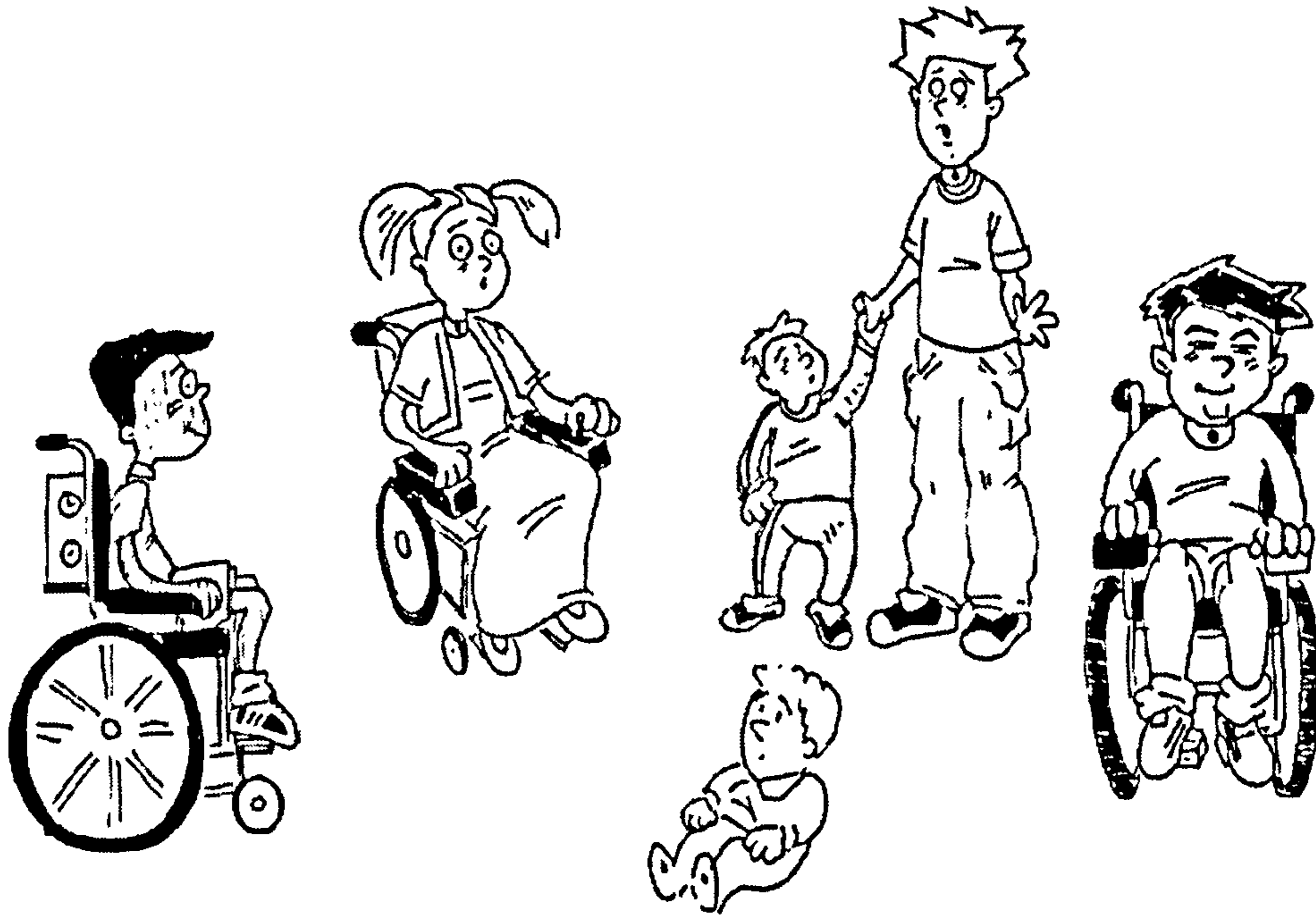
Education, leisure and cultural activities:

The child has a right to be educated, enter employment (Articles 23, 28, 29), and participate in socially integrated leisure and cultural activities (Articles 23, 31). This study found that:

- The young people were frequently deprived of an adequate education and access to extra curricular activities (especially in hospital);
- The young people generally found it difficult getting out and about and meeting friends. Because of this they did not always feel a valued part of society;
- The young people wanted to explore their sexuality and engage in relationships, and
- Some young people wanted to go to university and exercise their right to enter meaningful paid employment.

Appendix 3 - The CSRI Costing Instrument

Client Service Receipt Inventory (CSRI)



For Parents

**© Jane Noyes and the Personal Social Services Research Unit,
The University of Kent, UK.**

**Jane Noyes
Department of Health Sciences and Clinical Evaluation, University of York**

Client study number:

Date:
dd / mm / yy

Information gained from (state whom):
.....

Section 1 Sociodemographic Information

1.1 Child's date of birth *date*
dd / mm / yy

1.2 Child's age *years/months*
y m

1.3 Child's sex
1. female
2. male

1.4 Marital status (if child is over 16 yrs)
1. Single
2. Married
3. Cohabiting
4. Separated
5. Divorced
6. Other (describe) _____

1.5 Child's ethnic group _____
(refer to manual for groupings)

1.6 Child's country of birth _____
(refer to coding sheet)

1.7 Child's Mother tongue
1. National language
2. Other language (but having good knowledge of national language.)
3. Other language (and having poor or no knowledge of national language)
4. Pre-verbal child

1.8 Child's method of communication
1. Speech
2. Voice simulator
3. Non-verbal communication
4. Computer
5. Other (describe) _____

1.9 In your own words what is your child's diagnosis/diagnoses?

1.10 To date, how much time, in total, has your¹ child spent in hospital(s)?
yy / mm

Section 2 Your Child's *Current living situation*

2.1 In your own words what is your family profile?
(refer to manual for prompts)

2.2 Child's contact with parents

1. Regular contact with both parents
2. Regular contact with mother only
3. Regular contact with father only
4. Periodic contact with parent(s)
5. No contact with parents
6. Other (describe) _____

2.3 What type of accommodation does your child currently live in? (refer to manual for definitions)

Domestic / family

1. Owner occupied
2. Privately rented
3. Rented from local authority/housing assoc.

Hospital

4. Neonatal intensive care unit
5. Children's intensive care unit
6. Children's high dependency unit
7. Children's ward
8. Long term ventilation unit
9. Adult intensive care unit
10. Adult high dependency unit
11. Adult ward
12. Adult spinal injury unit

Community (non hospital)

13. Residential school
14. Staffed residential community for young disabled
15. Residential nursing home/care facility
16. Rehabilitation facility
17. Sheltered housing
18. Unstaffed group home
19. Foster care
20. Independent living scheme
21. Other (describe) _____

LAMINATE 1

2.4 What is your overall feeling regarding the appropriateness of where your child currently lives?

1. Completely Appropriate 2. Mostly Appropriate 3. Mixed 4. Mostly Inappropriate 5. Completely Inappropriate

Comments: _____

2.5 Has your child lived anywhere else in the last 12 months?

- 1. Yes
- 2. No
- 3. Not known

If yes: complete table:

Accommodation type (see Q 2.3 for code)	Number of days in last 12 months

2.6 Is your child 'looked after' by Social Services?

- 1. Yes
- 2. No
- 3. Not known

2.7 If your child currently lives in hospital or is 'looked after' by the local authority, have regular review meetings taken place under Section 85 of the Children Act (1989) in the last 12 months (refer to manual for definition)

- 1. All Review meetings took place as outlined in Section 85
- 2. Fewer review meetings took place than outlined in Section 85
- 3. No review meetings took place
- 4. Not known

Did your child attend Section 85 review meetings?

- 1. Attended all meetings
- 2. Attended some meetings
- 3. Too young
- 4. Not invited to attend
- 5. Invited but did not attend
- 6. Not known

Did you as parent(s) /guardians attend Section 85 reviews meetings?

- 1. Attended all meetings
- 2. Attended some meetings
- 3. Not invited to attend
- 4. Invited but did not attend
- 5. Not known

Comments: _____

Q 2.8 – 2.11 refer to children that are currently living in domestic/family accommodation.

2.8 If you currently live in domestic / family accommodation:

How many adults over the age of 18 years live there?

How many children under 18 years live there?

How many bedrooms are there? 3

2.9 What is your child's current living situation?

1. Living with parent(s) (+/- siblings)
2. Living with other relatives
4. Living with adoptive parents
5. Living with partner (+/- children)
6. Lives alone (with or without care team)
7. Other (describe) _____

2.10 What is the nature of your child's current domestic/family Accommodation?:

1. House (no adaptations)
2. House (with adaptations)
3. Flat (no adaptations)
4. Flat (with adaptations)
5. Bungalow (no adaptations)
6. Bungalow (with adaptations)
7. Purpose built (describe) _____
8. Other (describe) _____

LAMINATE 2

2.11 What areas of the accommodation have been adapted and are currently accessible to your child? (enter '0' if not relevant)

Area	Adaptations completed	Funded by*	Accessible to client 1=accessible, 2=not accessible	Additional work required
Drive				
Garage				
Front garden				
Front door				
Entrance hall				
Family kitchen				
Family dining room				
Family lounge				
Play room				
1 storey extension				
2 storey extension				
Loft conversion				
Stairs				
Client's Bedroom				
Client's living area				
Bathroom 1				
Bathroom 2				
Shower room 1				
Shower room 2				
Separate toilet 1				
Separate toilet 2				
Carer's lounge				
Carer's kitchen				
Back door				
Back garden				
Other				
Other				
Other				

* 1= NHS, 2= Social Services, 3= voluntary organisation, 4=private, 5= family.

LAMINATE 3

2.12 What is your overall feeling regarding your child's Domestic housing?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Comments: _____

LAMINATE 4

2.13 In the past 1 month what methods of transportation has your child used?
exclude journeys to school/college and ambulance transport
(enter '0' if service has not been used)

Transport	Provider*	Funded by*	Number of journeys
Your own car			
Family saloon car			
Family van/people carrier			
Black cab			
Mini cab			
Dial a ride			
Train			
Bus/coach			
Relatives (any vehicle)			
Friends (any vehicle)			
Other (describe)			

* 1= NHS, 2= Social Services, 3= voluntary organisation, 4=private, 5= parents/family/client, 6=other.

LAMINATE 5

2.14 What is the your overall feeling about the transportation available to your child?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Comments: _____

Section 3 Education and Training

Fill in questions 3.1 to 3.12 if your child is currently receiving education or training

- 3.1 Has your child a statement of educational need?
1. Yes
 2. No
 3. Not known

- LAMINATE 6
- 3.2 In the past 3 months of the school/college term what education services has your child used?
(Enter '0' if the service has not been used)

Service	Name of facility	Provider sector*	Funded by*	Mode of attendance/frequency
Hospital education service				
Mainstream primary school				
Special primary school				
Residential primary school (state type)				
Mainstream secondary school				
Special secondary school				
Residential secondary school (state type)				
Sixth form college				
College of further education (special needs)				
College of further education (mainstream)				
University				
Modern apprenticeship				
Home tuition by teacher				
Extra curricular lessons				
Other (describe)				
Other (describe)				

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family

- LAMINATE 7
- 3.3 In the past 3 months of the school/college term how has your child been transported to school/college? (Enter '0' if the service has not been used)

Transport	Provider sector*	Frequency of journeys	Distance to school
School bus			
Taxi			
Parents			
Other (describe)			
Other (describe)			

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family

LAMINATE 8

3.4 **In the past 3 months** of the school/college term what **additional** education services has your child used?
(Enter '0' if service not used)

Service	Number of contacts	Duration of contact (mins/hours)
School doctor		
Educational psychologist		
School nurse		
Education welfare officer		
Other (describe)		
Other (describe)		

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family

LAMINATE 9

3.5 **In the past 3 months** of the school /college term has your child had any **additional support in the classroom?** (Enter '0' if service not used)

Service	Provider sector*	Funded by*	Hours per week
Unqualified classroom assistant/helper			
Personal classroom teacher			
Transcribing / typing services			
Own personal carer(s)			
Break-time assistant			
Walkie-talkie			
Other (describe)			

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family.

LAMINATE 10

3.6 **In the past 12 months** of the school/college term what is your feeling about **your child's attendance** at school/college?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

3.7 **In the past 3 months** of the school/college term **how many days** has your child been **absent?**

Days absent

What factors have affected your child's **attendance** at school/college?

LAMINATE 11

3.8 What is your overall feeling about the **education services** that your child **currently** receives?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Comments:

3.9 Are there any additional education services that your child currently needs? If yes, describe:

Are there any current services that your child needs more of? If yes, describe:

Are there any current services that your child would like less off or no longer needs? If yes, describe:

3.10 Does your child have any educational needs that currently require addressing?

1. Yes
2. No
3. Not known

If yes, describe:

Q 3.11 and 3.12 are for children over 14 years
LAMINATE 12

3.11 What educational qualifications is your child currently working towards, or has been awarded? (Enter '0' if not relevant)

Qualification	Number of subjects currently studying	Number of subjects passed
NVQ Level 1 and 2		
NVQ Level 3 and 4		
GCSEs		
A levels		
Scottish Highers		
S levels		
OND		
HND		
City and Guilds		
Undergraduate degree		
Specific vocational training (describe)		
Other (describe)		
Other (describe)		

3.12 What would your child like to do when they leave school/college?
(eg: further education/job/career/ambition etc.)

Section 3a

Education and Training of Parents

LAMINATE 13

Tick all

applicable

Maternal education	No qualifications	
	NVQ Level 1 and 2	
	NVQ Level 3 and 4	
	GCSEs	
	A levels	
	Scottish Highers	
	S levels	
	OND	
	HND	
	City and Guilds	
	Undergraduate degree	
	Specific vocational training (describe)	
	Other (describe)	
	Other (describe)	

Tick all

appropriate

Paternal education	No qualifications	
	NVQ Level 1 and 2	
	NVQ Level 3 and 4	
	GCSEs	
	A levels	
	Scottish Highers	
	S levels	
	OND	
	HND	
	City and Guilds	
	Undergraduate degree	
	Specific vocational training (describe)	
	Other (describe)	
	Other (describe)	

Section 4 *Employment and income*

Questions 4.1 – 4.3 are for children over 16 years old

4.1 What is your child's employment status?

1. Paid or self employment
2. Voluntary work
3. Sheltered employment
4. Supported employment (New Deal, Modern apprenticeship, job training etc)
5. Unemployed and looking for work
6. Full time student
7. At home and not looking for work
8. Exempt through disability
9. Other (describe) _____

4.2 If child is employed: state occupation _____

How many hours per week does your child work? *Hours per week*

How many days has your child been absent from work within the last 3 months?
Days absent from work

What factors have affected your child's attendance at work?

4.3 *If your child is unemployed:*

Number of weeks unemployed
within the last 3 months* *Number of weeks*
Assume 13 weeks = 3 months

4.4 What is the employment status of you as parent(s) and if appropriate partner?
(Enter '0' if not applicable)

1. Paid or self employed
2. Voluntary work
3. Unemployed *mother*
4. Student *father*
5. Housewife/husband *partner*
6. Unpaid carer for client
7. Retired
8. Exempt through disability
9. Other _____
10. Not known

LAMINATE 14/15

4.5 Do you receive any state benefits? 1. Yes
(include benefits claimed by parents/guardian/partner/carer in respect of the child) 2. No

If yes: What benefits are received? (please tick all boxes that apply)

Attendance Allowance (AA)

Higher rate £55.30 per week
Lower rate £37.00 per week

Budgeting loans (social fund) if yes, state date and amount.....

Child benefit

Eldest child £15.50 per week
Lone parent with protected rights £17.55 per week.....
Other children £10.35

Child Maintenance bonus One off payment of £1000

Child Support Maintenance (amount varies according to circumstances)

Community Care Grant If yes, state date and amount.....

Council Tax benefit

Crisis loans (social fund) if yes, state date and amount

Disability Living Allowance (DLA)

Paid because you need looking after:
Higher rate £55.30 per week
Middle rate £37:00 per week.....
Lower rate £14.65 per week

Paid to help you get around:
Higher rate £38.65
Lower rate ££14.65

Disability Living Allowance for children under 16 years

Paid because your child needs looking after:
Higher rate £53.55 per week
Middle rate £35.80 per week.....
Lower rate £14.20 per week

Paid to help you get your child around:
Higher rate £37.40
Lower rate ££14.20

Disabled Person's Tax credit (age 16years and over)

Guardian's Allowance

Eldest child £11.35 per week

Other child £9.85 per week

Housing benefit

Incapacity benefit

Income Support

Plus disability premium

Plus severe disability premium

Income Support for residential accommodation

Invalid Care Allowance

Jobseeker's Allowance

Severe Disablement Allowance (age 16 and over)

Statutory Sick Pay

Travel costs to and from hospital

Working Families' Tax Credit

Other (describe)

Other (describe)

4.6 How much do you receive altogether in benefits per week?

£ _____

Q 4.7 refers to people who live in domestic/family or community (non-hospital) accommodation
LAMINATE 16

4.7 What is the income of your household per week from all sources before taxes and deductions? Note: a household is either 1 person living alone, or a group of people (who may or may not be related) living, or staying temporarily at the same address with common housekeeping.

A	£0 - £192	(£0 - £10,000 year)	<input type="checkbox"/>
B	£193 - £288	(£10,001 - £15,000 year)	<input type="checkbox"/>
C	£289 - £384	(£15,001 - £20,000 year)	<input type="checkbox"/>
D	£385 - £480	(£20,001 - £25,000 year)	<input type="checkbox"/>
E	£481 - £576	(£25,001 - £30,000 year)	<input type="checkbox"/>
F	£577 or more	(£30,001 or more per year)	<input type="checkbox"/>

LAMINATE 17

4.8 Does your household currently have any financial debts?

- A None
- B less than £10,000
- C £10,001- £15,000
- D £15,001 - £20,000
- E £20,001 - £25,000
- F £25,001 - £30,000
- G £30,001 or more.....

Comments:
.....
.....
.....
.....
.....
.....
.....
.....
.....

Section 5 Services Receipt

If unable to provide all of the information can they suggest an appropriate person to contact such as their case manager?

Name: _____ Tel: _____

5.1 What method and type of assisted ventilation does your child use now?

Intermittent positive pressure ventilation via:

1. Tracheostomy
2. Facemask
3. Endo tracheal tube
4. Nasal mask
5. Nasal pillows

Continuous positive pressure ventilation via:

6. Tracheostomy
7. Facemask
8. Endo tracheal tube
9. Nasal mask
10. Nasal pillows

Intermittent negative pressure ventilation via:

11. Chamber (iron lung)
12. Chamber (iron lung) with additional CPAP mask
13. Hayek jacket
14. Cuirass shell

Continuous negative pressure ventilation via:

15. Chamber (iron lung)
16. Hayek jacket
17. Cuirass shell

Other:

18. Unilateral phrenic nerve pacing alone
19. Bilateral phrenic nerve pacing alone
20. Unilateral phrenic nerve pacing in addition to one of the above (specify) _____
21. Bilateral phrenic nerve pacing in addition to one of the above (specify) _____
22. None of the above (please specify) _____

5.2 Time dependent on assisted ventilation per 24 hour day:

1. Intermittent (describe use) _____
2. 24 hour use

5.3 Make of main ventilator used (if known): _____

5.4 Does your child have a second back up ventilator?

1. Yes
2. No

If yes, state type _____

5.5 Approximate year when ventilator(s) obtained:

- 1. Main ventilator
- 2. Second ventilator
- 3. Third ventilator

yy

5.6 How is the current ventilator(s) funded?

- 1. Purchased by parents/family
- 2. Purchased by Health Authority
- 3. Purchased by Social Services
- 4. Purchased by NHS Trust
- 5. Purchased by Primary Care Trust
- 6. Purchased by a Charity or Voluntary Organisation
- 7. Leased by Health Authority
- 8. Leased by Social Services
- 9. Other (please state) _____
- 10. Combination of above (please state) _____
- 11. Not known

5.7 Is the ventilator(s) covered by a service contract?

- 1. Yes
- 2. No
- 3. Not known

LAMINATE 18

5.8 Other equipment currently used by your child: (Enter '0' if not relevant)

Equipment type	Quantity	Approx year when obtained	Funded by*	Service contract 1=yes, 2=no, 3=unknown
Pulse oximeter				
Transcutaneous oxygen monitor				
Transcutaneous CO2 monitor				
End tidal CO2 monitor				
Suction machine				
Apnoea alarm				
Oxygen cylinders				
Oxygen concentrator				
Liquid oxygen machine				
Humidifier				
Electric pump (nutrition)				
Ambu bag				
Electric wheelchair (standard)				
Electric wheelchair (+ventilator)				
Manual wheelchair				
Electricity back up generator				
Back up power packs				
Car battery to power ventilator				
Mobile phone				
Electric fan				
Electric hoist (in ceiling)				
Free standing hoist				
Other (describe)				
Other (describe)				
Other (describe)				
Other (describe)				
Other (describe)				

* 1=NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family.

LAMINATE 19

5.9 Does your child currently have any of the following?
(Enter '0' if not relevant)

Item	Quantity	Funded by*
Standard car seat		
Adapted car seat		
Standard buggy/pushchair		
Adapted buggy/pushchair		
Walking aid (describe)		
Standing frame		
Communication aid (describe)		
Working dog for the disabled		
Hospital style bed		
Other (describe)		

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family.

5.10 Does your child have any equipment that you consider to be unnecessary or no longer needed?

1. Yes

2. No

If yes, specify what equipment:

5.11 Does your child currently need any additional items of equipment?

1. Yes

2. No

If yes, specify what equipment:

LAMINATE 20

5.12 **In the last 1 month what disposable equipment and supplies has your child used?**
(Enter '0' if not used)

Item	Quantity used per month	Prescription or non prescription	Ordered by	Obtained from	Source of funding	How delivered to client?
Tracheostomy tubes						
Tracheostomy tapes						
Sterile water (trachy)						
Sterile water (humidifier)						
Ventilator tubing						
Contact gel						
Electrolyte solution						
Pulse oximeter probes						
Portable oxygen cylinders						
Nasogastric tubes						
Gastrostomy tubes						
Giving set (feed)						
Suction catheters						
Suction tubing						
Urinary catheters						
Urinary catheter bags						
Nappies						
Enteral (liquid) nutrition						
Other (describe)						
Other (describe)						
Misc: dressings/tape etc:						

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family

LAMINATE 21

5.13 **In the last 6 months what specific items of low volume disposable equipment has your child used?** (Enter '0' if not used)

Item	Quantity used per 6 months	Prescription or non prescription	Ordered by	Obtained from	Source of funding*	How delivered to client?
Masks						
Nasal pillows						
Emergency oxygen cylinders						
Other (describe)						
Other (describe)						
Other (describe)						
Other (describe)						
Other (describe)						
Other (describe)						

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family

5.14 **In the last 12 months have you encountered any problems regarding disposable equipment/supplies?**

1. Yes
2. No

If yes, specify: _____

LAMINATE 22

5.15 In the last 1 month what medicines has your child taken?

Name of drug	Prescription/non prescription	Dosage (if known)	Dosage frequency	Length of course
1.				
2.				
3.				
4.				
5.				
6.				
7.				
8.				

LAMINATE 23

5.16 In the last 6 months has your child taken any 'one off' medicines or courses of medicines (for example, antibiotics):

Name of drug	Prescription/non prescription	Dosage (if known)	Dosage frequency	Length of course
1.				
2.				
3.				
4.				
5.				
6.				
7.				
8.				

5.17 Does your child currently have a designated employed care team specifically to look after them?
 (Exclude here carers/assistants employed by the education authority)

- 1. Yes
- 2. No

If yes, how many hours of care is provided per week and when do carers provide care (24hours/day/night/weekends/school)?

If no, move on to Q 5.21.

LAMINATE 24

5.18 List below the current employed care team (including vacancies):

Type of carer (refer to manual for definitions)	Qualified or unqualified (refer to manual for definitions)	Hours worked per week	Provider sector*
1.			
2.			
3.			
4.			
5.			
6.			
7.			
8.			
9.			
10.			
11.			
12.			
13.			

* 1= NHS, 2= Social Services, 3= voluntary organisation, 4=private, 5= family.

5.19 In the last 12 months have you encountered any problems regarding your child's employed care team?

1. Yes
2. No

If yes, specify:

LAMINATE 25

5.20 What is your overall feeling about the effectiveness of the employed care team ?

1. Completely Effective 2. Mostly Effective 3. Mixed 4. Mostly Ineffective 5. Completely Ineffective

Additional comments:

5.21 In what ways has the employed care team contributed to your child's quality of life?
comments:

Q 5.22 – 5.25 for those people that live in domestic/family accommodation only

LAMINATE 26

5.22 In the last 3 months what additional paid domestic care services have you used?
(Enter '0' if service not used)

Service	Provider sector*	Total number of contacts	If applicable - average contact time (hours)
Home help			
Laundry			
Clinical waste disposal			
Domestic cleaning			
Other (describe)			
Other (describe)			
Other (describe)			
Other (describe)			

* 1= NHS, 2= Social Services, 3= voluntary organisation, 4=private, 5= family

LAMINATE 27

5.23 In the last 3 months what unpaid care services have you used?
(Enter '0' if service not used)

Service	Provided by*	Total number of contacts	If applicable - average contact time (hours)
Personal care (bathing/dressing)			
Shopping			
Companion/befriending			
Escort			
Child/sibling minding			
Housework			
Gardening			
Preparing meals			
Generally providing support			
Looking after pets			
Other (describe)			
Other (describe)			

* 1=voluntary organisation, 2= parent/guardian, 3=partner, 4=sibling, 5= extended family, 6=friends, 7=other

For all people

LAMINATE 28

5.24 In the past 12 months what 'respite care' services have you used?
(Enter '0' if service not used)

Service	Provider sector*	Total number of contacts	If applicable - average contact time (hours/days)
Hospital			
Hospice			
Residential facility			
Other (describe)			
Other (describe)			
Other (describe)			
Other (describe)			

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family, 7=friend

LAMINATE 29

5.25 In the last 12 months what is your overall feeling about the respite services your child received?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Additional comments: _____

5.26 In the past 12 months has your child's 'care package' been reviewed?

1. Yes
 2. No
 3. Not known

If yes, describe who undertook the review: _____

Who attended the review?

1. Parent(s)/guardian/partner only attended
 2. Only index child attended
 3. Parent(s)/guardian/partner and index child attended
 4. Not invited to attend
 5. Invited but did not attend

LAMINATE 30

5.27 In the last 12 months what in-patient services has your child received?
exclude respite care (Enter '0' if the service has not been used)

Service	Name of facility	Reason for admission*	Number of admissions	Total number of inpatient days
PICU				
Adult ICU				
Long term ventilation unit				
High dependency unit				
Spinal Injuries unit				
Children's ward				
Adolescent unit				
Adult ward				
Hospice				
Rehabilitation centre				
Residential facility				
Other (describe)				
Other (describe)				
Other (describe)				

*1= emergency admission for treatment, 2= planned admission for treatment, 3= planned routine check-up, 4=staffing problems, 5= equipment failure, 6=power failure, 7=existing in-patient, 8=other.

Laminate 31

5.28 In the last 12 months what hospital-based services has your child used (either as an existing in-patient or on an out-patient basis)?
(Enter '0' if service not used)

Service	Name of facility	Unit of measurement	Number of units received
Specialist doctor		Appointment	
Specialist doctor		Appointment	
Specialist doctor		Appointment	
Specialist doctor		Appointment	
Accident & emergency dept		Attendance	
Accident & emergency dept		Attendance	
Physiotherapist		Hours of treatment	
Hydrotherapy		Hours of treatment	
Rehabilitation		Hours of treatment	
Occupational therapist		Hours of treatment	
Communication/language		Hours of treatment	
Wheelchair clinic		Appointment	
Dietitian		Hours of treatment	
Speech therapist		Hours of treatment	
Psychiatrist		Hours of treatment	
Psychologist		Hours of treatment	
Family therapist		Hours of treatment	
Counsellor		Hours of treatment	
Chiropodist		Hours of treatment	
Orthodontist		Hours of treatment	
Dentist		Hours of treatment	
Ophthalmologist		Appointment	
Social worker		Appointment	
Other (describe)			
Other (describe)			

LAMINATE 32

5.29 In the last 12 months what paramedic /ambulance services has your child used?
(Enter '0' if service not used)

Service	No of occasions
Emergency ambulance	
Non-emergency ambulance	
Rapid response team (from A&E)	
Mobile children's intensive care ambulance and transfer team	
Helicopter and paramedic team	
Plane and paramedic team	
Other (describe)	

LAMINATE 33

5.30 In the past 3 months have you contacted any of the following services for telephone help/advice?
 (record subsequent contact/appointments in Q 5.30 below)
 (Enter '0' if service not used)

Service	Number of times
Rang Specialist doctor	
Rang GP	
Rang case manager	
Rang team leader	
Rang social worker	
Rang practice nurse	
Rang specialist nurse	
Rang hospital(s) ward	
Rang parents' support group	
Searched the internet	
Rang NHS Direct	
Other (describe)	
Other (describe)	

LAMINATE 34/35

5.31 In the past 3 months what community care services has your child used?
 Do not include services provided by staff in the accommodation facility or out-patient services provided by a hospital. (Enter '0' if service has not been used)

Service	Provider sector*	Total number of contacts	Where seen?		Average contact time (mins/hours)
			Home	Community facility	
General practitioner					
Community paediatrician					
Specialist health visitor					
Health visitor					
Social worker					
District nurse					
Nursery nurse					
Practice nurse					
Children's community nurse					
Case manager					
Team leader					
Physiotherapist					
Occupational therapist					
Speech therapist					
Chiropodist					
Outreach worker/family support					
Family counselling/therapy					
Counselling/therapy					
Psychologist					
Optician					
Dentist					
Independent advocate/visitor					
'Befriender'					
Other (describe)					
Other (describe)					
Other (describe)					

* 1= NHS, 2= Social Services, 3= voluntary organisation, 4=private

LAMINATE 36

5.32 In the past 3 months what alternative/complimentary therapies has your child used?
 (Enter '0' if service has not been used)

Service	Provider sector*	Funded by*	Total number of contacts	Average contact time(hours)
Conductive therapy (PORTAGE)				
Acupuncture				
Reflexology				
Massage				
Aromatherapy				
Herbalist				
Homeopathy				
Other (describe)				
Other (describe)				
Other (describe)				

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family, 7=friend

LAMINATE 37

5.32 In the last 3 months what day activity services has your child used? (Enter '0' if service has not been used)

Service	Name of facility	Funded by*	Number of attendances	Average duration of attendance
Social services play group				
Private play group				
Social services nursery				
Private nursery				
Voluntary organisation day activity facility				
NHS day activity facility				
Social services day activity facility				
Other (describe)				
Other (describe)				

* 1= NHS, 2= Social Services, 3= education authority, 4= voluntary organisation, 5=private, 6= family.

LAMINATE 38

5.34 In the last 12 months what is your overall feeling about the total amount of care and services your child received?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Additional comments: _____

5.35 Are there any additional services that your child currently needs? If yes, describe: _____

Are there any current services that your child needs more of? If yes, describe: _____

Are there any current services that your child would like less off or no longer needs? If yes, describe: _____

LAMINATE 39
5.36 What is your feeling about the overall co-ordination of all the care and services that your child receives?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Comments: _____

LAMINATE 40
5.37 Over the last 12 months how would you generally describe your child's health?

1. Very well 2. Mostly well 3. Mixed 4. Mostly unwell 5. Very unwell

Additional comments: _____

5.38 Does your child currently have any health needs that require addressing?

1. Yes
2. No
3. Not known

If yes, describe: _____

LAMINATE 41
5.39 Over the last 12 months how would you generally describe your child's social life?

1. Completely Satisfied 2. Mostly Satisfied 3. Mixed 4. Mostly Dissatisfied 5. Completely Dissatisfied

Additional comments: _____

5.40 Does your child currently have any social needs that require addressing?

1. Yes
2. No
3. Not known

If yes, describe: _____

5.41 For children aged 13 years and above have arrangements been discussed or put into place for when they transfer to adult services?

- 1. Yes
- 2. No
- 3. Not known

4. Already transferred to adult services

If yes, describe

Additional Comments:

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References

- Aday L., Aitkin M. and Wegener D. (1988) *Pediatric Home Care: Results of a National Evaluation of Programmes for Ventilator Assisted Children*. Pluribus Press, Chicago, USA.
- Aday L. and Wegener D. (1988) Home care for ventilator-assisted children: Implications for the children, their families, and health policy. *Children's Health Care* 17 (2):112-120.
- Alderson P. (1999) *Learning and Inclusion. Cleves School Experience*. Fulton Publishing.
- Alderson P. (2000) The importance of theories in health care. *British Medical Journal* 317:1007-10.
- Alderson P. and Goodey C. (1998a) Theories in healthcare and research: Theories of consent. *British Medical Journal* 317:1313-1315.
- Alderson P. and Goodey C. (1998b) *Enabling Education: Experiences in Special and Ordinary Schools*. Tufnell Publishing.
- Altman D. (1991) *Practical Statistics for Medical Research*. Chapman and Hall, London.
- Allen N., Simone J. and Wingenbach G. (1994) Families with a ventilator-assisted child: transitional issues. *Journal of Perinatology* 14:48-55.
- Alexander J. and Willems E. (1981) Quality of Life: Some measurement requirements. *Archives of Physical Medicine and Rehabilitation* 62(6):261-265.
- Ambrosio I., Woo M., Jansen M. and Keens T. (1998) Safety of hospitalised ventilator-dependent children outside of the intensive care unit. *Pediatrics* 101 (2):257-9.
- American Thoracic Society Consensus Statement (1990) Home mechanical ventilation. *American Review of Respiratory Disease* 141:258-259.
- Arblaster L, Conway J, Foreman A. and Hawtin M. (1998) *Achieving the Impossible. Interagency Collaboration to Address the Housing, Health and Social Care Needs of People Able to Live in Ordinary Housing*. The Policy Press, Bristol.
- Ashworth W. (1996) *Inquiry into the Care and Treatment of Nicholas Gelderd*. North West Regional Health Authority, Warrington.
- Aspinal F., Addington-Hall., Hughes R. and Higginson I. (2003) Using satisfaction to measure the quality of palliative care: a review of the literature. *Journal of Advanced Nursing* 42(4):324-339.

Atlas Ti™ (2002) Version 4.2. Scientific Software Development, Berlin.

Audit Commission (1993) *Children First: A Study of Hospital Services*. HMSO, London.

Audit commission (2001) *Going Places: Taking People to and from Education, Social Services and Healthcare*. Audit Commission, London.

Audit Commission (2002a) *Fully Equipped: The Provision of Equipment to Older or Disabled People by the NHS and Social Services in England and Wales*. Audit Commission, London.

Audit Commission (2002b) *Special Educational Needs: A Mainstream Issue*. Audit Commission, London.

Audit Commission (2002c) *Data Remember: Improving the Quality of Patient-Based Information in the NHS*. Audit Commission, London.

Audit Commission (2002d) *Recruitment and Retention – A Public Service Workforce for the 21st Century*. Audit Commission, London.

Audit Commission (2002e) *Procurement and Supply: A Review of National Findings*. Audit Commission, London.

Audit Commission (2003) *Services for Disabled Children: A Review of Services for Disabled Children and their Families*. Audit Commission, London.

Babinski Y. (1994) Roadblocks to homecare. *Caring* 13 (12):18-20, 22, 24.

Bach J., Intintola P., Alba A. and Holland I. (1992) The ventilator assisted individual. Cost analysis of institutionalisation v rehabilitation and in-patient management. *Chest* 101(1):26-30.

Bach J., Vega J., Majors J. and Friedman A. (2003) Spinal muscular atrophy type1: Quality of life. *American Journal of Physical Medicine and Rehabilitation* 82(2):137-142.

Baldwin S. (1985) *The Costs of Caring: Families with Disabled Children*. Routledge and Kegan Paul, London.

Baldwin S. and Carlisle J. (1994) *Social Support for Disabled Children and their Families: A Review of the Literature*. HMSO, London.

Baldwin-Myers A. and Oppenheimer E. (1996) Quality of life and quality of care data from a 7 year pilot project for home ventilator patients. *Journal of Ambulatory Care Management* 19(1):45-59.

Ball M. (1998) *School Inclusion: The School, the Family and the Community*. Joseph Rowntree Foundation, York.

- Barker I. (1996) Central funding and a central resource providing information are needed. *British Medical Journal* 313:940.
- Baumgardner D. and Burtea E. (1998) Quality of life in technology-dependent children receiving home care and their families: A qualitative study. *Wisconsin Medical Journal* 97:51-55.
- BBC News website (2002) Pope against 'useless' treatment of dying. http://news.bbc.co.uk/1/hi/english/world/europe/newsid_1890000/1890636.stm. Accessed March 2004.
- Beckett J. (1996) The long road home: the creation of the 'Katie Beckett Waiver'. *Exceptional Parent: Parenting Your Child With a Disability* 26(6):68-70.
- Beecham J. (1995) Collecting and Estimating Costs. In Knapp, M (Ed) *The Economic Evaluation of Mental Health Care* pp 60-82. Ashgate, Aldershot, UK.
- Beecham J. and Knapp M. (1992) Costing Psychiatric Interventions. In Thornicroft G., Brewin C. and Wing J. (Eds). *Measuring Mental Health Needs* pp163-183. Gaskell, London.
- Beecham J., O'Neill T. and Goodman R. (2001) Supporting young adults with hemiplegia: Services and costs. *Health and Social Care in the Community* 9(1):51-59.
- Beresford B. (1997) *Personal Accounts: Involving Disabled Children in Research*. Social Policy Research Unit, York.
- Beresford B. (2003) *The Community Equipment Needs of Disabled Children and their Families*. Research Works 2003/01, Social Policy Research Unit, University of York, UK.
- Beresford B., Sloper T. and Baldwin S. (1996) *What Works in Services for Families with a Disabled Child?* Barnardos, Ilford, UK.
- Beresford B. and Oldham C. (2000) *Making Homes Fit for Children: Working Together to Promote Change in the Lives of Disabled Children*. The Policy Press/Joseph Rowntree Foundation, Bristol/York, UK.
- Beresford B. and Oldham C. (2002) *Housing Matters: National Evidence Relating to Disabled Children and their Housing*. The Policy Press/Joseph Rowntree Foundation, Bristol/York, UK.
- Bevan M. (2002) *Housing and Disabled Children: The Art of the Possible*. The Policy Press, Bristol, UK.
- Blair A., Burg J., Foran J., Gibb H., Greenland S. and Morris R. (1995) Guidelines for application of meta-analysis in environmental epidemiology. *Regulatory Toxicology and Pharmacology* 22:189-97.

- Bolton A. (1997) *Losing the Thread: Pupils' and Parents' Voices about Education for Sick Children*. National Association for the Education of Sick Children, London.
- Bradshaw J. and Mayhew E. (2004) Public attitudes to dependency and the welfare state. *International Journal of Market Research* 46(1):49-63.
- British National Formulary (March 2002) British Medical Association and the Royal Pharmaceutical Society of Great Britain, London.
- Brookes D., Tonack M., King A., Simson H. and Goldstein R. (2002) Ventilator users' perspectives on important elements of health-related quality of life. Unpublished.
- Brunner E. (1997) Socio-economic determinants of health: Stress and biology of inequality. *British Medical Journal* 314:1472-8.
- Brust J., Leonard B. and Sielaff B. (1992) Maternal time and the care of disabled children. *Public Health Nursing* 9(3):177-184.
- Bullinger M., Mackensen S. and Kirchberger I. (1994) KINDL – ein Fragebogen zur gesundheitsbezogenen Lebensqualität von Kindern. *Zeitschrift für Gesundheitspsychologie* 2:64-67.
- Bullinger M., Schmidt S, Peterson C. and the DISABKIDS Group (2002). Assessing quality of life of children with chronic health conditions and disabilities: a European approach. *International Journal of Rehabilitation Research* 25:197-206.
- Bywaters P., Zoebial A., Qulsom F., Wallace L. and Singh G. (2003) Attitudes towards disability amongst Pakistani and Bangladeshi parents of disabled children in the UK: Considerations for service providers and the disability movement. *Health and Social Care in the Community* 11(6):502-509.
- Byford S. and Rafferty J. (1998) Perspectives in economic evaluation. *British Medical Journal* 316:1529-1530.
- Canlas-Yamsuan M., Sanchez I., Kesselman M. and Chernick V. (1993) Morbidity and mortality patterns of ventilator-dependent children in a home care programme. *Clinical Pediatrics* 32:706-713.
- Campbell J. (2003) Choose Life: There is growing support for moves to help severely disabled people to die. But why not help them instead? *The Guardian*, 26th August:28
- Carers UK (2002) *Without Us? Calculating the Value of Carers' Support*. Carers UK, London.
- Capen C. and Dedlow E. (1998) Discharging ventilator-dependent children: A continuing challenge. *Journal of Pediatric Nursing* 13(3):175-84.

CASP (Critical Appraisal Skills Programme). (2002) 10 Questions to make sense of qualitative research. CASP, UK.

Children Act (1989) HMSO, London.

Coffman S. (1995) Crossing Lines: Parents' experiences with paediatric nurses in the home. *Rehabilitation Nursing Research* 4(4):136-43.

Cohen M. (1999) The technology-dependent child and the socially marginalized family: A provisional framework. *Qualitative Health Research* 9(5):654-688.

Colville G. and Mok Q. (2003) Psychological management of 2 cases of self-injury on the paediatric intensive care unit. *Archives of Disease in Childhood* 88(4):335-6.

Community Care (Direct Payments) Act (1996), HMSO, London.

Conservative Party (2003) *Setting the NHS Free: A Conservative Policy Consultation*. Conservative Party Central Office, London.

Copeland L. and Clements D. (1993) Parental Perceptions and Support Strategies in Caring for a Child with a Chronic Condition, *Issues in Comprehensive Pediatric Medicine* 16:109-121.

Corser N. (1996) Sleep of 1 and 2 year old children in intensive care. *Issues in Comprehensive Pediatric Nursing* 19:17-31.

Curren A., Sharples P., White C. and Knapp M. (2001) Time costs of caring for children with and without disabilities. *Developmental Medicine and Child Neurology* 43(8):529-33.

Dalton R. and Kirkhart K. (1985) An evolution of emotional problems faced by ventilator-assisted children. *The Psychiatric Forum* 13:73-81.

Darbyshire P. (1996) *Living with a Sick Child in Hospital. The Experiences of Parents and Nurses*. Chapman & Hall, London.

Davies R. (1996) A difficult case: Home ventilation of a child with motor and sensory neuropathy. *British Medical Journal* 313:153-154.

Denzin N. and Riessman C. (1995) Narrative analysis. *Journal of Communication* 45:177-184.

Department of Culture Media and Sport (2003) *Beyond 2004: A Framework for Action on Disability*. DCMS, London.

Department for Education and Science, and Department of Health (2003) *Together from the Start: Practical Guidance for Professionals Working with Disabled Children (birth to third birthday) and their Families*. Department for Education and Science, and Department of Health, London.

Department for Education and Science (2003) *Special Education Needs Action Programme*. Department for Education and Science, London.

Department of Health (1996) *The Patient's Charter. Services for Children and Young People*. HMSO, London.

Department of Health (1997a) *Paediatric Intensive Care 'A Framework for the Future'. Report From the National Co-ordinating Group on Paediatric Intensive Care to the Chief Executive of the NHS Executive*. Department of Health, London.

Department of Health (1997b) *A Bridge to the Future. Nursing Standards, Education and Workforce Planning in Paediatric Intensive Care. Report of the Chief Nursing Officer's Task Force*. Department of Health, London.

Department of Health (1998a) *The Quality Protects Programme. Transforming Children's Services*. Department of Health, London.

Department of Health (1998b) *Objectives for Social Services for Children*. Department of Health, London.

Department of Health (1998c) *Unlocking the Potential: Effective Partnerships for Improving Health*. Department of Health, London.

Department of Health (1998d) *Partnership in Action (New Opportunities of Joint Working Between Health and Social Services)*. Department of Health, London.

Department of Health (1998e) *A Guide to Receiving Direct Payments*. Department of Health, London.

Department of Health (1999a) *Discharging Responsibilities, Discharge Liaison Nurses and Their Role in Discharge Planning*. Department of Health, London.

Department of Health (1999b) *Direct Payments: Cash for Services. Information Video*. Department of Health, London.

Department of Health. (2000a) *The NHS Plan: A Plan for Investment, a Plan for Reform*. Department of Health, London.

Department of Health. (2000b) *For the Benefits of Patients: A Concordat with the Private and Voluntary Health Care Provider Sector*. Department of Health, London.

Department of Health (2000c) *Community Care (Direct Payments) Act: Policy and Practice Guidance 2nd Edition*. Department of Health, London.

Department of Health (2000d) *An Easy Guide to Direct Payments*. Department of Health, London.

Department of Health (2001a) *Guide to Integrating Community Equipment Services*. Department of Health, London.

Department of Health (2001b) *Valuing People. A New Strategy for Learning Difficulties for the 21st Century*. The Stationery Office, London.

Department of Health (2002a) *NHS Finance Manual*. Department of Health, London.

Department of Health (2002b). *Growing Capacity: A New Role for External Healthcare Providers in England*. Department of Health, London.

Department of Health (2002c) *Listening, Hearing and Responding. Department of Health Action Plan: Core Principles for the Involvement of Children and Young People*. Department of Health, London.

Department of Health (2002d) *Diagnosis Coding ICD-10. Introduction to the International Statistical Classification of Diseases and Related Health Problems. Tenth Revision*. Department of Health, London.

Department of Health (2002e) *NHS National IT Modernisation Programme: Delivering 21st Century Support for the NHS*. Department of Health, London.

Department of Health (2003a) *Getting the Right Start: The National Service Framework for Children, Young People and Maternity Services. Emerging Findings*. Department of Health, London.

Department of Health (2003b) *Green Paper: Every Child Matters*. HMSO, London.

Department of Health (2003c) *Listening, Hearing and Responding: Department of Health Involving Children and Young People*. Department of Health, London.

Department of Health (2003d) *National Service Framework for Children of Wales. Template for the Development of Standards*. Department of Health, London.

Department of Health, Department for Education and Employment, Home Office. (2000) *Framework for the Assessment of Children in Need and their Families*. The Stationery Office, London.

DeWitt P., Janson M. and Davidson Ward S. (1993) Obstacles to discharge of ventilator-dependent children from the hospital to the home. *Chest* 103:1560-1565.

Dhillon J., Frewen T., Singh N. and Speechley K. (1996) Chronic mechanical ventilation-dependent children in Canada. *Paediatrics and Child Health* 1:111-6.

Diehl S., Moffitt K. and Wade S. (1991) Focus group interview with parents of children with medically complex needs: An intimate look at their perceptions and feelings. *Children's Health Care* 20(3):170-178.

Disability Discrimination Act (1995) HMSO, London.

- Dixon-Woods M., Agarwal S., Young B., Jones D. and Sutton A. (2004) *Integrative Approaches to Qualitative and Quantitative Evidence*. NHS Health Development Agency, London.
- Dobson B. and Middleton S. (1998) *Paying to Care: The Cost of Childhood Disability*. York Publishing Services, York.
- Doyal L. and Gough I. (1991) *A Theory Of Human Need*. Macmillan, London.
- Drummond M., O'Brien B., Stoddart G. and Torrance G. (1997) *Methods for the Economic Evaluation of Health Care Programmes*. Second Edition. 1997. Oxford University Press, Oxford.
- Drummond M. and Jefferson T. (1996) Guidelines for authors and peer reviewers of economic submissions to the BMJ. *British Medical Journal* 313:275-283.
- Dyer C. (2000) Boy paralysed in road accident to get £5 million. *The Guardian* March 23rd: 9.
- Dyer C. (2002) Woman makes legal history in right to die case. *British Medical Journal* 324:629.
- Dyer C. (2003) Mother to challenge ventilator decision: Disabled girl's treatment may become test case. *The Guardian* 12th July:7.
- Edenbrook D., Stevens V., Hibbert C., Mann A. and Wilson A. (1997) A new method of accurately identifying costs of individual patients in intensive care: the initial results. *Intensive Care Medicine* 23:645-50.
- Eiser C and Morse R. (2001) *Quality of Life Measures in Chronic Diseases of Childhood*. Health Technology Assessment 5(4).
- Effey A. (1999) Time analysis: social contacts in the course of days in a child treated with ventilator assistance in an intensive care unit. *Pneumologie* 53 Supplement 2: S98-9.
- EuroQol Group (1998) *EuroQol EQ-5D: User Guide*. EuroQol Group, University of York, UK.
- EuroQol Group (1990) EuroQol: A new facility for the measurement of health-related quality of life. *Health Policy* 16:199-208.
- Evans D. (2002) Systematic reviews of interpretative research: interpretative data synthesis of processed data. *Australian Journal of Advanced Nursing* 20:22-26.
- Fauroux B., Howard P. and Muir J. (1994) Home treatment for chronic respiratory insufficiency: the situation in Europe. The European working group

on home treatment for chronic respiratory insufficiency. *European Respiratory Journal* 7:1721-6.

Fazil Q., Bywaters P., Ali Z., Wallace L. and Singh G. (2002) Disadvantage and discrimination compounded: The experience of Pakistani and Bangladeshi parents of disabled children in the UK. *Disability and Society* 17(3):237-253.

Felinas D., O'Connor P. and Miller R. (1998) Quality of life for ventilator-dependent ALS patients and their caregivers. *Journal of the Neurological Sciences* 160 (supplement 1) S134-136.

Fields A., Rosenblatt A., Pollock M. and Kaufman J. (1991) Home care cost-effectiveness for respiratory technology-dependent children. *American Journal of Diseases in Childhood* 145, 729-33.

Fischer A. (1989) Long-term management of the ventilator-dependent patient: levels of disability and resocialisation. *European Respiratory Journal* 2, supplement 7:651-654.

Fleming J., Chanella M. and Eland J. (1994) Impact on the family of children who are technology dependent and cared for in the home. *Pediatric Nursing* 20:370-388.

Flesch R. (1918) A new readability yardstick. *Journal of Applied Psychology* 32:221-3.

Foot M. (1973) *Aneurin Bevan*. Cavis-Poynter, London.

Frates R., Splaingard M., Smith E. and Harrison G. (1985) Outcome of home mechanical ventilation in children. *The Journal of Pediatrics* 106(4):850-856.

Fraser J., Mok Q. and Tasker R. (1997) Survey of occupancy of paediatric intensive care units by children who are dependent on ventilators. *British Medical Journal* 315:347-348.

Fraser J., Henrichsen T., Mok Q. and Tasker R. (1998) Prolonged mechanical ventilation as a consequence of acute illness. *Archives of Disease in Childhood* 78(3):253-6.

Fulop N., Allen P., Clarke A. and Black N. (2003) From health technology assessment to research on the organisation and delivery of health services: addressing the balance. *Health Policy* 63:155-165.

Gardner B., Theocleous F., Watt J. and Krishnan K. (1985) Ventilation or dignified death for patients with high tetraplegia. *British Medical Journal* 291:1620-1622.

Gilgoff I. and Helgren J. (1992) Planning an outing from hospital for ventilator-dependent children. *Developmental Medicine and Child Neurology* 43(10):904-10.

- Gilgoff R. and Gilgoff I. (2003) Long-term follow-up of home mechanical ventilation in young children with spinal cord injury and neuromuscular conditions. *The Journal of Pediatrics* 142(5):476-80.
- Gillick v West Norfolk and Wisbech Area Health Authority* (1985) 3 All ER 402, 421, 423.
- Gipson T., Sivak E. and Gullede D. (1987) Psychological aspects of ventilator dependency. *Psychiatric Medicine* 5:245- 255.
- Glass v United Kingdom* (2004) no. 61827/00, ECHR.
- Glendinning C. and Kirk S. (1998) Trends in community care and patient communication: Implications for the roles of informal carers and community nurses in the United Kingdom, *Journal of Advanced Nursing* 38 (2):370-381.
- Glennerster H. (2003) *Understanding the Finance of Welfare: What Welfare Costs and How to Pay for it*. The Policy Press, Bristol.
- Goldberg A., Faure E., Vaughn C., Snarski R. and Seleny F. (1984) Home care of life supported persons: An approach to program development. *The Journal of Pediatrics* 10(5):785-795.
- Gorman C., Kennedy P. and Hamilton L. (1998) Alterations in self-perceptions following childhood onset of spinal cord injury. *Spinal Cord* 36:181-185.
- Gough D., and Elbourne D. (2002) Systematic research synthesis to inform policy, practice and democratic debate. *Social Policy and Society* 1:225-236.
- Granger A. (2001). Chapter 2: Systematic review of child health-related quality of life measures. In *Health-related quality of life after paediatric intensive care: Development and validation of a package of outcome measures*. Unpublished PhD thesis, University of York, UK.
- Greco V., Sloper P. and Barton K. (2004) *Care Co-ordination and Key Worker Services for Disabled Children in the UK*. Research Works 2004-01, Social Policy Research Unit, University of York, UK.
- Gross D. (1984) Relationships at risk: issues and interventions with a disturbed mother-infant dyad. *Perspectives in Psychiatric Care* 22(4):159-64.
- Guyatt G., Feeny D. and Patrick D. (1993) Measuring health-related quality of life. *Annals of Internal Medicine* 118:622-9.
- Hall S. and Nicoll A. (1998) The British Paediatric Surveillance Unit: A pioneering method for investigating the less common disorders of childhood. Report of a seminar held in June 1995. *Child: Care, Health and Development* 24(2):129-143.
- Hadden F. (1996) Home ventilation of a child with motor and sensory neuropathy: Life does have a price and the rest of us have to pay for it. *British Medical Journal* 313: 940.

- Hammer J. (2000) Home mechanical ventilation in children: indications and practical aspects. *Schweiz Medical Wochenschr* 130(49):1894-902.
- Harris J. (1987) QALYfying the value of life. *Journal of Medical Ethics* 13:117-23.
- Hazlett D. (1989) A study of paediatric home ventilator management: medical, psychosocial, and financial aspects. *Journal of Paediatric Nursing* 4(4):284-294.
- Heaney L., McAllister D. and MacMahon J. (1999) Cost minimisation of provision of oxygen at home: Are the drug tariff guidelines cost effective. *British Medical Journal* 319:19-23.
- Health Act* (1999) HMSO, London.
- Heaton J., Noyes J., Sloper P, and Shah R. (2003) *Technology and Time: Home Care Regimes and Technology-Dependent Children*. Final Report. ESRC, London.
- Helgeson V. (2003) Social support and quality of life. *Quality of Life Research* 12(1):25-31.
- Hill S. and Harris U. (1993) The outcomes process: some reflections from research with people in their 60s and 70s. *Critical Public Health* 4(4): 21-28.
- Heidegger M. (1962) *Being and Time*. Blackwell, Oxford.
- Heywood F. (1996) *Funding Adaptations. The Need to Co-operate*. The Policy Press, Bristol.
- Heywood F. (2001) *Money Spent Well: The Effectiveness and Value of Housing Adaptation*. The Policy Press/ Joseph Rowntree Foundation, Bristol/York, UK.
- Holloway I. (1997) *Basic Concepts for Qualitative Research*. Blackwell Science, Oxford.
- HM Treasury (2000) *HM Spending Review: New Public Spending Plans 2003-6*. HM Treasury, London.
- Human Rights Act* (England) (1998). HMSO, London.
- James I. (1996) Centralised paediatric intensive care beds are blocked. *British Medical Journal* 312:1426.
- Janowski M. (1984) Accidental disconnections from breathing systems. *American Journal of Nursing* (February): 241- 244.
- Jardine E. and Wallis C. (1998) Core guidelines for the discharge home of the child on long-term assisted ventilation in the United Kingdom. *Thorax* 53:762-767.

- Jardine E., O'Toole M., Payton J. and Wallis C. (1999) Current status of long-term ventilation of children in the United Kingdom: Questionnaire survey. *British Medical Journal* 318:295-299.
- Jenkinson C., Stradling J. and Peterson S. (1997) Comparison of three measures of quality of life outcome in the evaluation of continuous positive airways pressure therapy for sleep apnoea. *European Sleep Research Society* 6:199-204.
- Kagan C., Lewis S. and Heaton P. (1998) *Caring to Work: Accounts of Working Parents of Disabled Children*. Family Policy Studies Centre, London.
- Kamm M., Burger R., Rimensberger P., Knoblauch A. and Hammer J. (2001) Survey of children supported by long-term mechanical ventilation in Switzerland. *Swiss Medical Weekly* 131:261-266.
- Kaufman S. (2003) Hidden places, uncommon persons. *Social Science and Medicine* 56:2249-2261.
- Kemper K. (1994) Is this hospitalisation really necessary? *Contemporary Pediatrics* 11:43-56.
- Kennedy I. (1982) Reflections on the Arthur trial. *New Society* 7th January: 13-15.
- Kind P., Dolan P., Gudex C. and Williams A. (1998) Variations in population health status: results from a United Kingdom national survey. *British Medical Journal* 316:736-41.
- Kirk S. (1998) Families' experiences of caring at home for a technology dependent child: A review of the literature. *Child: Care, Health and Development* 24(2):101-114.
- Kirk S. (1999) Caring for children with specialised health care needs in the community: the challenges for primary care. *Health and Social Care in the Community* 7 (5):350-357.
- Kirschbaum M. (1996) Life support decisions for children: What do parents value? *Advances in Nursing Science* 19(1):51-71.
- Knapp M. (1993) Background Theory. In Netten A. and Beecham J. (eds) *Costing Community Care Theory and Practice* pp 9-24. Ashgate, Aldershot, UK.
- Knight A. (1998) *Valued or Forgotten? Disabled Children and Independent Visitors*. National Children's Bureau, London.
- Krishnan K. (1993) Recommendations concerning the costing and management for patients requiring domiciliary ventilation. *International Medical Society of Paraplegia* 31:276-279.

- Landgraf J., Maunsell E., Speechley K., Bullinger M., Campbell S. and Abetz L. (1998) Canadian-French, German and UK versions of the Child Health Questionnaire: Methodology and preliminary item scaling results. *Quality of Life Research* 7(5):433-45.
- LaPlante M. and Kaye H. (1998) *Trends in Disability and Their Causes: Proceedings of the Fourth National Disability and Statistics and Policy Forum*. Washington DC, US Department of Education.
- Lawford J. and Eiser C. (2001) Exploring links between the concepts of quality of life and resilience. *Pediatric Rehabilitation* 4(4):209-16.
- Leonard, B., Brust, J. and Nelson, R. (1993) Parental Distress: Caring For Medically Fragile Children At Home. *Journal of Pediatric Nursing* 8(1):22-30.
- Lenton S., Margolan H., Fraser J. and Simms P. (2001) Service and financial planning for long term ventilation in childhood-prevalence, predictions and NHS costs. Unpublished.
- Lightfoot J. and Sloper P. (2002) *Having a Say in Health: Guidelines for Involving Young Patients in Health Services Development*. Social Policy Research Unit, University of York, UK.
- Lindahl B., Sandman P. and Rasmussen B. (2003) Meanings of living at home on a ventilator. *Nursing Inquiry* 10(1):19-27.
- Local Government Act* (1999) HMSO, London.
- Locker D. and Dunt D. (1978) Theoretical and methodological issues in sociological studies of consumer satisfaction with medical care. *Social Science and Medicine* 12:283-292.
- Ludvigsen A. and Morrison J. (2003) *Breathing Space: Community Support for Children on Long-term Ventilation*. Barnardos, London.
- Lumeng J., Warschausky S., Nelson V. and Augenstein K. (2001). The quality of life of ventilator-assisted children. *Pediatric Rehabilitation* 4(1):21-27.
- Lynch M. (1990) Home care of the ventilator-dependent child. *Children's Health Care* 19(3):169- 173.
- Lyth I. (1982) *The Psychological Welfare of Children Making Long Stays in Hospital: An Experience in the Art of the Possible*. The Tavistock Institute of Human Relations, London.
- Macduff C. and Russell E. (1998) The problem of measuring change in individual health-related quality of life by postal questionnaire: use of the patient-generated index in a disabled population. *Quality of Life Research* 7(8):761-9.

- Mahon A., Glendinning C., Clarke K. and Craig G. (1996) Researching children: Methods and ethics. *Children and Society* 10:145-154.
- Margolan H. (2001) *Bristol Royal Hospital: Project Report – To Investigate the Needs of Long Term Ventilated Children and their Families in the South West Region*. United Bristol Healthcare Trust, Bristol.
- Maynard F. and Munth A. (1987) The choice to end life as a ventilator-dependent quadriplegic. *Archives of Physical Medicine and Rehabilitation* 68(12):862-4.
- Marks D. (1997) Models of disability. *Disability and Rehabilitation* 19(3): 85-91.
- McGinley M., Baus E., Gyza K., Johnson K., Lipton S., Magee M., Moore F. and Wojtyak D. (1996) Multidisciplinary discharge planning: Development of a process. *Nursing Management* 27(10):55-60.
- McKeevor B. (2002) *Homes Fit for Children: From Policy to Politics*. Guildhall Press, Derry, Northern Ireland.
- McMahon S. (2003) *Scottish Economic Report: Household Debt in Scotland and the United Kingdom*. Scottish Executive, Edinburgh.
- McWilliam C. and Wong C. (1994) Keeping it secret: The costs and benefits of nursing's hidden work in discharging patients. *Journal of Advanced Nursing* 19(1):152-163.
- Medical Law Monitor (1998) Resuscitation of terminally ill children: Re C (A Minor). *Medical Law Monitor* 5(2):1-2.
- Microsoft (2000) Microsoft Corporation, USA.
- Ministry of Health (1959) *Report of the Committee on the Welfare of Children in Hospital (Platt Report)*. HMSO, London.
- Morse J. and Field P. (1996) *Nursing Research: The Application of Qualitative Approaches*. (Second edition) Chapman and Hall, London.
- Morris J. (1998a) *Don't Leave Us Out. Involving Disabled Children and Young Children with Communication Impairments*. York Publishing Services Ltd. York.
- Morris J. (1998b) *Still Missing? Volume 1. The Experiences of Disabled Children and Young People Living Away from their Families. The Who Cares? Trust*. London.
- Morris J. (1998c) *Still Missing? Volume 2. Disabled Children and the Children Act*. The Who Cares? Trust. London.
- Motwani J. and Herring G. (1988) Home care for ventilator-dependent persons: a cost effective public policy. *Health and Social Work* 13:20-24.

Muckherjee, S., Beresford B. and Sloper P. (1999) *Unlocking Key Working: An Analysis and Evaluation of Key Worker Services for Families of Disabled Children*. The Policy Press/Joseph Rowntree Foundation, Bristol/York.

Murphy K. (1997) Parenting a technology assisted infant: coping with occupational stress. *Social Work and Health Care* 24(3-4):113-26.

Murphy E., Dingwall R., Greatbatch D., Parker S. and Watson P. (1998) Qualitative research methods in health technology assessment: a review of the literature. *Health Technology Assessment* 2 (16).

N v Department of Health In Northern Ireland. (2003) Unpublished deposition to the Court.

National Assembly for Wales (2000) *Children and Young People: A Framework for Partnership*. National Assembly for Wales.

National Children's Bureau (2003) *Guidelines for Research*. National Children's Bureau, London.

National Hospital for Neuro-disability (2002) *'Life Long Home' Ventilator Unit at Putney: 2002 prices*. NHND, London.

Netten A. and Curtis L. (2002) *Unit Costs of Health and Social Care*. Personal Social Services Research Unit, University of Kent.

NHS Executive North West. (2000) *A Specification for the Provision of Paediatric Long-term Ventilation for the Area Covered by the NHS Executive North West*. NHS Executive North West, Warrington United Kingdom.

NHS Executive and Department of Health (2000) *Evaluation of the Powered Wheelchair and Voucher Scheme Initiatives. Final Report*. York Health Economics Consortium, University of York. UK

Noreau L. and Shepherd R. (1995) Spinal cord injury, exercise and quality of life. *Sports Medicine* 20(4):226-250.

North and South Thames PICU Network (2002) *Draft Guidelines for the Development of Services for Children who Require Long-term Ventilation*. North and South Thames PICU Network, London.

Nosek M. and Holmes A. (1996) Ventilator users: Independent living and quality of life. *Physical Medicine and Rehabilitation Clinics of North America* 7(2): 445-456.

Noyes J., Hartmann H., Samuels M. and Southall D. (1999) The experiences and views of parents who care for ventilator-dependent children. *Journal of Clinical Nursing* 8:440-450.

- Noyes J. (1999a) *Voices and Choices. Young People Who Use Assisted Ventilation: Their Health, Social Care, and Education*. The Stationery Office, London.
- Noyes J. (1999b) Life as a Ventilator-dependent Child. *Journal of Social Work Practice* 13(2): 177-190.
- Noyes J. (2000a) 'Ventilator-dependent' children who spend prolonged periods of time in intensive care units when they no longer have a medical need or want to be there. *Journal of Clinical Nursing* 9:774-783.
- Noyes J (2000b). Are nurses respecting and upholding the human rights of children and young people in their care? *Paediatric Nursing* 12 (2):23-27.
- Noyes J. (2000c) Enabling young 'ventilator-dependent' people to express their views and experiences of their care in hospital. *Journal of Advanced Nursing* 31(5):1206-15.
- Noyes J. (2002) Barriers that delay children and young people who are dependent on mechanical ventilators from being discharged from hospital. *Journal of Clinical Nursing* 11(1):2-11.
- NSPCC (1997) *Turning Points*, NSPCC, London.
- O'Brien M. (2001) Living in a house of cards: family experiences with long-term childhood technology dependence. *Journal of Pediatric Nursing* 16(1):13-22.
- O'Brien M. and Wegner C. (2002) Rearing the child who is technology dependent: perceptions of parents and home care nurses. *Journal of Specialised Pediatric Nursing* 7(1): 7-15.
- Oldman C. and Beresford B. (1998a) *Homes Unfit for Children. Housing, Disabled Children and their Families*. The Policy Press, Bristol.
- Oldman C. and Beresford B. (1998b) *Living at Home with a Disabled Child*. The Policy Press, Bristol.
- Oliver M. (1998) Theories of disability in health practice and research. *British Medical Journal* 317:1446-9.
- Office of National Statistics (2001) *Living in Britain: Results from the 2001 General Household Survey*. Government Statistical Service. The Stationery Office, London.
- Office of National Statistics (2002 release) *The British Household Panel Survey (BHPS)*. Government Statistical Service. The Stationery Office, London.
- Office of National Statistics (2002 release) *The 1970 Birth Cohort Study*. Government Statistical Service. The Stationery Office, London.
- Office of National Statistics (2002 release) *UK Labour Force Surveys*. Government Statistical Service. The Stationery Office, London.

- Orlowski J. (1993) Ethical and quality of life issues in ventilator-dependent children. *Clinical Pediatrics* 32:714-717.
- Paditz E., Zieger S. and Bickhardt J. (2000) Self-reported quality of life in home mechanical ventilation in childhood, adolescence and young adulthood: Differences between parents and children. *Somnologie* 4:13-19.
- Palfrey J., Walker D., Haynie M., Singer J., Porter S., Bushey B. and Cooperman P. (1991). Technology's children: Report of a State-wide census of children dependent on medical supports. *Pediatrics* 87(5):611-618.
- Palmer S. and Raftery J. (1999) Opportunity cost. *British Medical Journal* 318:1551-2.
- Patterson J., Leonard B. and Titus J. (1992) Home care for medically fragile children: impact on family health and well-being. *Developmental and Behavioural Pediatrics* 13(4):248-255.
- Perrin J. (2002) Health services research for children with disabilities. *The Milbank Quarterly* 80(2):303-324.
- Pickin C. and St. Leger S. (1993) *Assessing Health Need Using the Lifecycle Framework*. OU Press, Buckingham.
- Pilmer S. (1994) Prolonged mechanical ventilation in children. *Pediatric Clinics of North America* 41(3):473-512.
- Piskule A. and Johnson P. (1997) Role of the interdisciplinary team function in the successful discharge of a ventilator-dependent person with a spinal cord injury. *Psychosocial Process* 10(2):67-72.
- Prasad R. (2003) Loyal cause: A pioneering scheme that provides a dedicated nurse for disabled people admitted to hospital. *The Guardian, Society*, 3rd September: 160.
- Pretty v Department of Public Prosecutions* (2001) European Court Human Rights, Admin 788.
- Puddicome B. (1995) *Face to Face: Communicating with People who do not use Language*. Values into Action, London.
- Purtilo R. (1986) Ethical issues in the treatment of chronic ventilator-dependent patients. *Archives of Physical Medicine and Rehabilitation* 67:718-721.
- Quint R., Chesterman E., Crain L., Winkleby M. and Boyce T. (1990) Home care for ventilator-dependent children: Psychosocial impact on the family. *American Journal of Disease in Childhood* 144:1238-1241.
- R v North West Lancashire Health Authority* (2000) 1 WRL 977.

- Ray L. and Ritchie J. (1993) Caring for chronically ill children at home: Factors that influence parents' coping. *Journal of Pediatric Nursing* 8(4):217-225.
- Raulin A. and Shannon K. (1986) Pediatric nurse practitioners: Case managers for technology dependent children. *Pediatric Nursing* 12:338-340.
- Ravens-Sieberer U. and Bullinger M. (1998) Assessing the health-related quality of life in chronically ill children with the German KINDL: First psychometric and content-analytical results. *Quality of Life Research* 4(7):399-407.
- Ravens-Sieberer U. and Bullinger M. (2000) *KINDL: Questionnaire for Measuring Health Related Quality of Life in Children and Adolescents. Revised Version. Manual.* KINDL, Germany.
- Reeve C. (1998) *Still Me.* Century, London.
- Rennick J. (1995) The changing profile of acute childhood illness: a need for the development of family nursing knowledge. *Journal of Advanced Nursing* 22:258-266.
- Richardson M., Student E., Boyle D., Smyth M. and Wheeler T. (1992) Establishment of a State-supported, specialized home care program for children with complex health-care needs. *Issues In Comprehensive Pediatric Nursing* 15:93-122.
- Ritchie J. and Spencer L. (1995) Qualitative data analysis for applied policy research pp 173-194. In Bryman A. and Burgess R. (Eds) *Analysing Qualitative Data.* Routledge, London.
- Roberts K. and Lawton D. (2001) Acknowledging the extra care parents give their disabled children. *Child: Care, Health and Development* 27(4):307-319.
- Robertson J. (1958) *Young Children in Hospital.* Tavistock, London.
- Robinson R. (1990) Ventilator dependency in the United Kingdom. *Archives of Disease in Childhood* 65:1235-6.
- Rogerson R. (1995) Environmental and health-related quality of life: conceptual and methodological similarities. *Social Science and Medicine* 41(10):1373-1382.
- Rosenbaum S. (1992) Mothers and children last: the Oregon Medicaid experiment. *American Journal of Law and Medicine* 18(1 and 2): 97-126.
- Rothwell P., McDowell Z., Wong C. and Dorman P. (1997) Doctors and patients don't agree: cross sectional study of patients' and doctors' perceptions and assessments of disability in multiple sclerosis. *British Medical Journal* 314:1580-4.

Royal College of Paediatrics and Child Health (1997) *Withholding or Withdrawing Life Saving Treatment in Children: A Framework for Practice*. RCPCH, London.

Ruggeri M. (1994) Patients' and relatives satisfaction with psychiatric services: the state of the art and its measurement. *Social Psychiatry and Psychiatric Epidemiology* 29(5):212- 227.

Ruta D., Garratt A., Leng M., Russell I. And MacDonald L. (1994) A new approach to the measurement of quality of life. The Patient-Generated Index. *Medical Care* 32(11):1109-26.

Sakakihara Y., Yamanaka T., Kajii M. and Kamoshita S. (1996) Long-term ventilator assisted children in Japan: a national survey. *Acta Paediatrica Japan* 38:137-42.

Samuels M. (1996) Long-term ventilation. *Paediatric Respiratory Medicine* 2: 24-31.

Sasaki S., Sugai K., Fukumizu M., Hanaoka S. and Makiko K. (2001) Mechanical ventilation care in severe childhood neurological disorders. *Brain and Development* 23:796-800.

Schwerdt M. (1996) Growing up on the intensive care unit. Results of a survey on quality of life of long-term ventilated children on intensive care units. *Medizinische Klinik Supplement* 2:53-5.

Sennhauser, F (1997) Quality of life in chronic respiratory disorders in children. *European Respiratory Review* 7(42):77-81.

Sevick M., Kamlet M., Hoffman L. and Rawson I. (1996) Economic cost of home based care for ventilator-assisted individuals. *Chest* 109(6):1597-1606.

Shiple L. (1997) Technology-dependent children at home. *Nursing in Critical Care* 2(5):235-238.

Simmonds A., Ward S., Heather S., Bush A. and Muntoni F. (2000) Outcome of paediatric domiciliary mask ventilation in neuromuscular and skeletal disease. *European Respiratory Journal* 16:476-481.

Smyth M. and Robus N. (1989) *The Financial Circumstances of Families with Disabled Children Living in Private Households*. OPCS Surveys of Disability in Great Britain Report 5. HMSO, London.

Social Policy Research Unit (SPRU) (2000a) *Introducing an Outcome Focus Into Care Management and User Surveys*. Research Works. November 2000. SPRU, University of York, UK.

Social Policy Research Unit (SPRU) (2000b) *Implementing an Outcomes Approach to Carer Assessment and Review*. Research Works. November 2000. SPRU, University of York, UK.

Social Policy Research Unit (SPRU) (2000c) *Surveying Outcomes of Equipment and Adaptations*. Research Works. November 2000. SPRU, University of York, UK.

Sokol D. (2003). To live and let die: In many US hospitals clinical ethicists are constantly on call. We should follow suit. *The Guardian* 11th September.

Sørli V., Lindseth A., Udén G. and Norberg A. (2000) Women physicians' narratives about being in ethically difficult care situations in paediatrics. *Nursing Ethics* 7(1):47-62.

Sørli V., Førde R., Lindseth A. and Norberg A. (2001) Male physicians' narratives about being in ethically difficult care situations in paediatrics. *Social Science and Medicine* 53:657-667.

Sørli V., Jansson L. and Norberg A. (2003) The meaning of being in ethically difficult care situations in paediatric care as narrated by female registered nurses. *Scandinavian Journal of Caring Science* 17(3):285-92.

Southall D., Cronin B., Hartmann H., Harrison-Sewell C. and Samuels M. (1993) Invasive procedures in children receiving intensive care. *British Medical Journal* 306:1512-3.

Special Education Needs and Disability Act (2001) HMSO, London

Spence A. (1995) Home ventilation: how to plan for discharge. *Nursing Standard* 9(42):38-40.

Sperber A. (2004) Translation and validation of study instruments for cross-cultural research. *Gastroenterology* 126(1 Supplement 1): S124-8.

SPSS TM (2001) version 11. SPSS Incorporated, USA.

Sritippayawan S., Kun S., Keens T. and Davison-Ward S. (2003) Initiation of home mechanical ventilation in children with neuromuscular diseases. *The Journal of Pediatrics* 142(5):481-485.

Stalker K., Carpenter J., Connors C. and Philips R. (2004) Difficulties gaining access to children in hospital for social research. In press *Child: Care, Health and Development*.

Steward M., Bussey K., Goodman G. and Saywitz K. (1993) Implications of developmental research for interviewing children. *Child Abuse and Neglect* 17 (1):25-37.

Sudbery J. and Noyes J. (1999) *The Voices and Choices of Young People who use Assisted Ventilation: Bibliography and Analysis of the Literature*. Joseph Rowntree Foundation/University of Salford, Manchester, UK.

Sudman S., Sirkin M. and Cowan C. (1988) Sampling rare and elusive populations. *Science* 240:991-996.

- Teague B., Fleming J., Castle A., Kiernan B., Lobo M., Riggs S. and Wolfe J. (1993) High tech home care for children with chronic health conditions: A pilot study. *Journal of Pediatric Nursing* 8:226-232.
- Tichy A., Braam C., Meyer T. and Rattan N. (1988) Stressors in pediatric intensive care units. *Pediatric Nursing* 14:40-42.
- Thompson R. (1990) Vulnerability in research: A developmental perspective on research risk. *Child Development* 61:1-16.
- Thyen U., Terres N., Yazdgerdi S. and Perrin J. (1998) Impact of long-term care of children assisted by technology on maternal health. *Developmental and Behavioural Pediatrics* 19(4): 273-282.
- Torgerson D. and Spencer A. (1996) Marginal costs and benefits. *British Medical Journal* 312:35-36.
- Torgerson D. and Raftery J. (1999) Measuring outcome in economic evaluations. *British Medical Journal* 318:1413.
- Townsley R., Abbott D. and Watson D. (2004) *Making a Difference? Exploring the Impact of Multi-Agency Working on Disabled Children with Complex Health Care Needs, Their Families and the Professionals who Support Them*. The Policy Press, Bristol, UK.
- Turnball H., Summers J., Poston D. and Beegle G. (2000) *Enhancing Family Quality of Life Through Partnerships and Core Concepts of Disability Policy*. Paper presented to the 11th International Association of the Scientific Study of International Disability World Congress, 29th-31st July, Seattle, WA, US.
- United Nations (1989) *United Nations Convention on the Rights of the Child. Including: Background to the Convention and Full Text of the Convention*. Children's Rights Development Unit, London.
- U.S. Department of Health and Human Services. (1988) *Task Force on Technology Dependent Children. Fostering Home and Community-Based care for Technology dependent Children*. Department of Health and Human Services, Washington, D.C.
- U.S. Office of Technology Assessment. (1987) *Technology-dependent Children: Hospital v Home care - A Technical Memorandum*. US Government Printing Office, Washington D.C.
- Vick S. (1996) Caring for ventilated patients in the community: A pilot study examining costs, quality of life and preferences. *Health and Social Care in the Community* 4(6):330-337.
- Ward L. (1997) *Seen and Heard: Involving Disabled Children and Young People in Research and Development Projects*. York Publishing Services Ltd. York.

- Warner J. and Norwood S. (1991) Psychosocial concerns of the ventilator-dependent child in pediatric intensive care units. *Clinical issues in Critical Care Nursing* 2(3):432-445.
- Warnock M. (2002) Why patients need the right to die: Miss B had no obligation to stay alive just to satisfy her doctors. *The Guardian*, Friday 22nd March.
- Weeks W. and Wallace A. (2002) Readability of British and American medical prose at the start of the 21st century. *British Medical Journal* 325:1451-2.
- Wilson S., Morse J. and Penrod J. (1998) Absolute involvement: the experience of mothers of ventilator-dependent children. *Health and Social Care in the Community* 6(4):224-233.
- Williams G. (1981) Downs Syndrome and the duty to preserve life. *New Law Journal* 8th October: 1040-1.
- Williams R. and Wright J. (1998) Epidemiological issues in health needs assessment. *British Medical Journal* 316:1379-1382.
- World Health Organisation (1958) *Constitution of the World Health Organisation. Annex 1. In: The first 10 years of the World Health Organisation.* WHO Geneva.
- World Health Organisation (1980) *International Classification of Impairments, Disabilities and Handicaps.* World Health Organisation, Geneva.
- Wright J. and Williams R. (1998) The development and importance of health needs assessment. *British Medical Journal* 316:1310-1313.
- Wright J., Williams R. and Wilkinson J. (1998) Development and importance of health needs assessment. *British Medical Journal* 316:1310-13.
- Yaksic J., DeWoody S. and Campbell S. (1996) Case management of chronic ventilator patients: Reduce average length of stay and cost by half. *Nursing Case Management* 1(1):2-10.