Health-Related Quality of Life (HRQoL) after Paediatric Intensive Care (PIC): Development and Validation of a Package of Outcome Measures

Angela Ruth Grange MA RSCN/RGN

A thesis submitted as fulfilment for the degree of Doctor of Philosophy

University of York

Department of Health Sciences

September 2002

Abstract

Paediatric intensive care (PIC) is expensive, providing high-technology medicine, where future prospects of survival and quality of life are uncertain. Outcome measurement in PIC is unsatisfactory and recognised as a national research priority. Subjective multidimensional, health-related quality of life (HRQoL) outcome measures may amplify data obtained from morbidity and mortality statistics by describing the quality not the quantity of survival. They may be of value in comparing outcomes in clinical trials, evaluating interventions, commissioning care programmes, assessing outcomes of new treatments, and contributing the patient's viewpoint; assisting policy-makers to improve the effectiveness of existing healthcare services.

This thesis makes two contributions to the evidence base for outcome measurement in PIC. First, a systematic review and evaluation of published child HRQoL outcome measures and synthesis of the evidence relating to the development and psychometric properties, user-centredness, clinical utility and feasibility of the measures applied in PIC. Secondly, the development of the first PIC, system-specific (respiratory) measure of HRQoL, the Paediatric Intensive Care Quality of Life (PICQoL) questionnaire, for use in a UK PIC population.

The systematic review describes limited availability of child HRQoL measures and information on their psychometric properties. Evaluation of these measures indicated that no measure adequately fulfilled the required measurement properties to evaluate specific dimensions of health in children less than five years of age, requiring PIC for a respiratory illness. The development and validation of a new HRQoL measure for use in this PIC population is described. Initial evidence suggests the measure has satisfactory psychometric properties, but further research is required to test its construct validity, responsiveness, and reliability in other PIC settings. The measure may provide a mechanism for evaluating the effectiveness of PIC. The role of evaluative HRQoL measures in routine clinical practice, research, clinical decision-making and patient management warrants further investigation in PIC settings.

Contents **Page**

Contents (continued) **Page**

Contents (continued) **Page**

Contents (continued) **Page**

Contents (continued) **Page**

Contents (continued) **Page**

Contents (continued) **Page**

Contents (continued) **Page**

Contents (continued) **Page**

List of tables **Page**

List of tables (continued) **Page**

List of tables (continued) **Page**

List of tables (continued) **Page**

List of figures **Page**

Preface

I have been practising as a paediatric nurse for twelve years, and I have received specialist training in caring for critically ill children and their families. My interest in investigating this research topic stemmed from the desire to find out more information about the quality of survival for children that my colleagues and I cared for on the paediatric intensive care unit (PICU), Leeds General Infirmary. I knew from my own clinical experience that many children survived PICU admission but I did not know at what cost this was to them and their families. Was the care that my colleagues and I delivered on the PICU effective? The focus of this thesis is the development and validation of a health-related quality of life (HRQoL) outcomes measure for use in children following PIC. This thesis describes an account of my research programme and experiences.

The opportunity to undertake the research programme described in this thesis arose via a three-year, full-time regional NHS research training fellowship. The research programme described was devised for the fellowship application. The fellowship also provided the opportunity to enhance my research knowledge and skills through formal research training by pursuing doctoral studies. The final year of my doctoral studies was completed part-time with a need to return to clinical practice on the PICU at Leeds General Infirmary to fund the final year. In the future, I hope to pursue a research career within an NHS setting, and to further develop the HRQoL measure described in this thesis.

Acknowledgement

I am grateful to a large number of people for their help and support throughout my doctoral studies. In particular, I wish to acknowledge the NHS Northern and Yorkshire Regional Research and Development office for funding my research programme via their regional fellowship scheme. I am also grateful to my employers, the Leeds Teaching Hospitals NHS Trust, for their support, in particular Michelle Milner (PICU nursing team leader) and Dr Mark Darowski (PICU lead clinician).

I am especially grateful to my supervisors Hilary Bekker (University of Leeds, formerly University of York) and Ian Russell (University of York), for their time and expertise, and constant support, guidance, and encouragement. Thanks also to Andrew Garratt (University of Oxford, formerly University of York) for supervising the first year of my research programme and guiding my fellowship application along with Ian Russell. I am

also grateful to members of my research advisory group (RAG): Daphne Russell for her statistical support; and Dr Mark Darowski, Dr Simon Whiteley, Dr Philip Holland, and Professor Malcolm Levene for providing a clinical perspective to my research programme. Thanks also to Jane Noyes (University of York) for her help with the systematic review; Gareth Parry and Chris McCabe (University of Sheffield) for their time and expertise regarding the use of the HUI measure; and Jeanne Landgraf (HealthcareAct, Boston) for her advice on scale development. Thanks also to Ellie Theile and Helen Matusewich (family care sisters on PICU) for providing support to parents who required family care intervention during and following my research programme; and to Debbie Doyle (former PICU audit clerk) for assisting with data reliability checks.

Special thanks to my husband Martin, for his unfailing support and tolerance, and encouragement, during the last four years; and for proof reading this manuscript along with Kate Hill. Special thanks also to my family, friends, and all the parents and clinicians who participated in my research study. Finally, I wish to acknowledge my uncle, Dr David Stevens FRCP (Lond), FRCPH, DCH and my late grandfather, Mr Edgar Stevens FRCS (Ed and Lond). Both influenced me to pursue a career in nursing in the NHS and I would like to dedicate this thesis to them both.

Author's declaration

I have worked on this research programme with support from my supervisors and members of my RAG. I took lead responsibility for developing the research protocol, day-to-day management of the research, patient recruitment, data collection, data management, statistical analysis and report writing. Jane Noyes reviewed a sample of papers in the systematic review as a second reviewer to check the reliability of my data extraction form and review procedure. Hilary Bekker performed the coding of two parental interviews in Phase I to assess the inter-rater reliability of my coding procedures. Some items were adapted from the Child Health Questionnaire, with the permission of the developer Jeanne Landgraf. The thesis has been composed by me and has not been included in any previous application for a degree. All sources of information have been acknowledged; errors and omissions in the text are my own.

Angela Ruth Grange

September 2002

CHAPTER 1 – Outcome measurement in paediatric intensive care (PIC)

1.0 Introduction

This thesis presents a three-phase research programme conducted in a UK regional paediatric intensive care unit (PICU) to develop and validate a health-related quality of life (HRQoL) measure for use in children under the age of five years, admitted to a PICU with a respiratory illness. This introductory chapter provides an overview of the issues relating to the measurement of HRQoL outcomes in the context of health policy, paediatrics, and paediatric intensive care (PIC). The current evidence will be summarised and gaps identified. Subsequent chapters will describe a systematic review of child HRQoL measures; the development of parent-generated and clinician-generated items for the HRQoL measure; the testing of the items for importance, agreement and dimensionality; and the testing of the reliability and validity of the HRQoL measure in a PIC population. The thesis concludes with a discussion chapter incorporating recommendations for future research.

1.1 Background

1.1.1 Health policy context

The purpose of the NHS is to improve the physical and mental health of the people of England (NHSE, 1996a). The World Health Organisation (WHO) defines health as "a state of complete physical, mental, and social wellbeing, and not merely the absence of disease or infirmity" (WHO, 1948). It is recognised that in order to improve health decisions about the provision and delivery of clinical services, they need to be driven increasingly by evidence of clinical and cost-effectiveness, coupled with systematic assessment of actual health outcomes (NHSE, 1996a). Care provided should be appropriate to people's needs, effective – drawing on the best available clinical evidence, and efficient and economic – to maximise health gain for the population (DoH, 1998). The evidence is clear that effective care improves clinical outcomes and that comparative evidence can stimulate improvements in efficiency (Baker, 1998).

Clinical effectiveness provides a framework for promoting evidence-based practice to improve quality in health care (Baker, 1998). It has been described as the extent to which specific clinical interventions, when deployed in the field for a particular patient or population, do what they are intended to do – i.e. maintain and improve health and secure

the greatest possible health gain from the available resources (including available evidence) (NHSE, 1996a). Evidence-based practice is the conscientious, explicit, and judicious use of current best evidence, based on systematic review of all available evidence – including patient-reported, clinician-observed and research-derived evidence – in making and carrying out decisions about the care of individual patients (Cullum, 1998).

The NHS Health Technology Assessment (HTA) programme is one NHS based initiative aiming to ensure high quality research information on costs, effectiveness and broader impact of health technologies is produced in the most efficient way for those who use, manage and work in the NHS (National Co-ordinating Centre for Health Technology Assessment, 1997). Health services research within this programme tests hypotheses and generates new knowledge that may subsequently be useful in improving the effectiveness or efficiency of health care (Russell, 1996). This research is important not just to researchers, but patients, managers, and clinicians; if we do not know how best to assess the advantages and disadvantages of treatments, their cost, and impact, then informed judgements – about individual treatment choices and about societal rationing decisions – are impossible (Stevens, Milne & Black, 1998). Health services research provides evidence on evaluation of interventions, including the development of generic outcome measures and patient-assessed outcomes, and includes secondary research in which primary studies of different types are synthesised to enhance their value to clinicians and other health service decision makers (Stevens et al, 1998).

The effectiveness of health care interventions and services can be assessed and monitored through the process of clinical and economic evaluation. In the context of a health service, evaluation is defined as the critical assessment, on as an objective basis as possible, of the degree to which entire services or their component parts (e.g. diagnostic tests, treatments, caring procedures) fulfil stated goals (St Leger, Schnieden & Walsworth-Bell, 1994). Economic evaluation is defined as the comparative analysis of alternative courses of action in terms of both their costs and consequences to provide information that will assist decision-makers to determine the most efficient way of allocating their scarce resources between competing demands (Drummond et al, 1997; Brazier & Deverill, 1998).

Randomised-controlled trials (RCTs) provide a 'gold standard' method for evaluating clinical effectiveness; it has become common practice for economic evaluations to be

conducted alongside these clinical evaluations (RCTs) and other studies of effectiveness (Brazier & Deverill, 1998). However, few RCTs generally report an economic evaluation (Prescott et al, 1998). Often the measurement of clinical outcomes, considered to be important to patients, is neglected owing to the limited availability of suitable outcome measures. More measures are needed to evaluate outcomes of health care and treatment from the patient perspective, including subjective measures of HRQoL. The demand for subjective outcome measurements of health and HRQoL has accelerated owing to the rise of clinical epidemiology and the emphasis on evidence-based medicine, which has increased the questioning of the effectiveness of many established medical treatments (Bowling, 1995; McDowell & Jenkinson, 1996).

1.1.2 Quality of life (QoL) and health-related quality of life (HRQoL)

The WHO definition of health has been highly influential in defining the quality of life (QoL) construct, i.e. disease state and physical symptoms, functional status, psychological functioning, and social functioning (Ware, 1984). The concept has been described as broad ranging, and affected in a complex way by a person's physical health, psychological state, level of independence, social relationships, and their relationships to salient features of the environment (WHOQOL Group, 1993). Spilker (1996) also adds economic and/or vocational status and factors, and religious and/or spiritual status to the definition. It is also recognised that an individuals' QoL is shaped by the culture and value systems in which they live (Cheater, 1998). Quality of life has been described as having four important properties: it is multifactorial, it is patient self-administered, its value is variable over time, and it is subjective (Schipper, Clinch & Olweny, 1996).

The concept of HRQoL was initially developed with adult illness populations and refers to the specific impact of an illness or injury, medical treatment, or health care policy on an individual's QoL (Patrick & Bergner, 1990; Speith & Harris, 1996). It is a multidimensional construct covering physical, emotional, mental, social and behavioural components of wellbeing and function as perceived by patients or observers, to capture the full range of manifestations of health and illness (Vivier, Bernier & Starfield, 1994; Bullinger & Ravens-Sieberer, 1995). Patrick and Berger (1990) describe HRQoL as encompassing five broad categories of concepts, including duration of life, impairments, functional status, health perceptions, and opportunities. Health-related quality of life has also been described as including functional status, physiologic status, perceptions of

wellbeing, and general life satisfaction (MacKeigan & Pathak, 1992). Others have included spiritual and role functioning dimensions in their definition (Spilker & Revicki, 1996). Paediatric HRQoL has been defined as encompassing the concepts of disease state, functional status, psychological functioning and social functioning (Speith & Harris, 1996). The term HRQoL is described as differing from QoL in that HRQoL is often used to exclude certain aspects of life, such as income, freedom, quality of the environment, that do not directly affect health issues (Guyatt, Feeny & Patrick, 1993).

1.1.3 The purpose of HRQoL outcome measures

There are three primary purposes for measuring HRQoL outcomes: discrimination, evaluation and prediction (Guyatt et al, 1993). Discriminative HRQoL measures differentiate between people who have a better HRQoL. Evaluative HRQoL measures identify longitudinal changes in HRQoL within patients during a period of time, and may be used for determining treatment benefit in clinical trials (Haley et al, 1991; Guyatt et al, 1993). They can be used to complement clinical evaluation by health care professionals by providing a broader picture of patient functioning from the patient perspective; a large body of evidence demonstrates that health care professionals are poor judges of how aspects of life quality are affected by illness and treatment for individual patients (Jenkinson & McGee, 1998). Evaluation of patient HRQoL and health preferences may also guide health care professionals and patients to select the treatment option which is most suited to the individual. Health-related quality of life measures, such as utility measures, which assess quality-adjusted-life years (QALYs), may also be used in resource allocation considerations (Jenkinson & McGee, 1998). Predictive HRQoL measures can be used to screen patients needing particular care or attention (Jenkinson & McGee, 1998). Screening measures must be able to identify both individuals who exhibit the phenomenon in question (test sensitivity) and those who do not (test specificity); they can identify needs for particular services and identify appropriate interventions (Jenkinson & McGee, 1998). Data about HRQoL in children with chronic conditions, compared to those without, are important for health care planning, and for evaluating the impact of different treatments and cultures on HRQoL in children generally (Ravens-Sieberer & Bullinger, 1997).

1.1.4 Types of HRQoL measure

Different types of HRQoL measures exist and have been described as generic or specific measures (Guyatt et al, 1993; Fitzpatrick et al, 1998a). Generic HRQoL measures have the

ability to detect HRQoL changes across different diseases and changes arising from organ systems other than the one that is the target of treatment (MacKeigan & Pathak, 1992). This is important when patients have multiple chronic conditions or when there are adverse effects of treatment. They are commonly developed for descriptive epidemiological or social science research applications, and to evaluate types of care or patient management (McDowell & Newell, 1996). If we want to know the HRQoL of a child compared with the normal healthy population of similar-aged children, generic measures are preferable (Eiser & Morse, 2001a). Generic HRQoL measures, however, are not exhaustive in their assessment and additional disease-specific or domain-specific information may be required under certain conditions (Gemke & Bonsel, 1996). Generic measures may not be responsive to small changes in children's conditions and a disease-specific measure may be more capable of detecting subtle improvements resulting from treatment, because it includes only those elements that are most important to the disease (MacKeigan & Pathak, 1992; Eiser & Morse, 2001a).

Specific measures can be specific to a disease, population, function or problem (Guyatt et al, 1993), and are generally designed for clinical application and to be sensitive to change following treatment - responsiveness (MacKeigan & Pathak, 1992; McDowell & Newell, 1996). They have the advantage of reducing patient burden and increasing acceptability by including only the relevant dimensions (Fletcher et al, 1992). Disadvantages include the lack of comparability of results with those from other disease groups and the possibility of missing effects in dimensions that are not included (Fletcher et al, 1992). Regarding their use in paediatric populations, some children may also have more than one condition in which it may be necessary for the child, or proxy, to complete multiple disease-specific measures (Eiser & Morse, 2001a). It is recommended that both generic and specific measures be administered as a package of outcome measures in QoL trials to minimise the limitations of both types of measure (Fletcher et al, 1992; MacKeigan & Pathak, 1992). The specific measure produces evidence most relevant to the clinician and is also most responsive to the main effects of an intervention, while a generic measure produces information relevant to a broader policy community (including those requiring comparisons across interventions and disease groups) and may also detect unexpected positive or negative effects of a novel intervention (Fitzpatrick et al, 1998a).

Health-related quality of life measures may also be classified according to the level at which they aggregate scores on separate dimensions: health profiles, health indices, preference-based measures, or battery measures (MacKeigan & Pathak, 1992; Eiser & Morse, 2001a). Health profiles, such as the SF-36 (Ware & Sherbourne, 1992), measure HRQoL across a number of distinct dimensions and include multiple items, which are grouped into different dimensions of functioning; they can be used in most populations (Eiser & Morse, 2001a). The dimensions of health and functioning represented in the profiles are given as separate scores, and can also be presented as a single aggregate score across the dimensions (Fletcher et al, 1992; MacKeigan & Pathak, 1992). Health indices, such as the EuroQoL (EuroQoL Group, 1990) measure HRQoL via a single summary score, and can be used in cost-benefit analysis – such single indices must include social valuations of health states (Jenkinson & McGee, 1998). Preference-based measures may involve the direct assessment of preferences for health states often concentrating on producing single-score scales using a utility approach; or the use of multi-attribute health status classification systems, producing scores for each health state (Eiser & Morse, 2001a). Utility measures, such as the Health Utilities Index (HUI) (Torrance et al, 1995), take account of the values attached to individual HRQoL states through explicit weighting, usually represented by a single score along a continuum of 0 (death) to 1 (perfect health) (MacKeigan & Pathak, 1992). These scores can be used to calculate quality-adjusted-life-years (QALYs) which are used in economic evaluations of health care and cost-utility analysis (MacKeigan & Pathak, 1992). A health battery is a collection of independent measures or individual dimensions from different measures that have been assembled to obtain a comprehensive HRQoL assessment; scores are reported separately for each measure or dimension (MacKeigan & Pathak, 1992; Eiser & Morse, 2001a).

1.1.5 Measurement issues in paediatric HRQoL assessment

The development of child health status and HRQoL measures has lagged behind the development of similar measures for adults partly owing to the conceptual and methodological complexities of developing measures in children (Schor, 1998). Morbidity and mortality is less prevalent in the paediatric age group as compared with adults, so that different types of measures relevant to child health are needed (Vivier et al, 1994).

Difficulties arise in deciding which dimensions of function to include and exclude; often what constitutes 'QoL' in children is decided from an adult perspective and based on the

conceptualisations of the investigator (Rosenbaum & Saigal, 1996). To measure HRQoL in children there must be a consensus as to what constitutes 'health' in this population. Child health has been defined as the ability to participate fully in developmentally appropriate activities requiring physical, psychological, and social energy (Pantell & Lewis, 1987). There is no general agreement regarding the normal roles and functions of children, nor predictable linear patterns of development available or few norms established with respect to age (Stein & Jessop. 1981; Haley et al, 1991; Jenney, Kane & Lurie, 1995). A child's health and health behaviour is also sensitive to the social context in which he or she lives; the family has a significant impact on a child's health through genetic and familial predispositions, learned health beliefs and values, and shared physical, social and emotional environments (Stein & Jessop, 1990; Schor, 1995 & 1998). It is also difficult to establish universal criteria because of the social and cultural differences across groups of children (Marra et al, 1996).

Measurement of health over time is more desirable than a single point or short-term measure, but is particularly difficult to assess in children as we expect them to progress in growth and development, but not in a predictably linear fashion (Pantell & Lewis, 1987; Starfield, 1987; Rosenbaum, Cadman & Kripalami, 1990). The normal incremental improvement in children's skills makes it difficult to know whether a particular treatment has truly had an effect (Schor, 1998). Assessing the HRQoL of children has unique challenges because of the varying developmental capabilities of children at different ages (Rosenbaum & Saigal, 1996). It may be difficult to determine whether the failure of a child to achieve an area of independent function reflects an abnormal development process, relationship with an environment that fosters dependency, or loss of ability to function because of illness (Stein & Jessop, 1990). The dimensions of interest incorporated in the measures used with younger children may not be applicable for older children because of the acquisition of more advanced skills (Saigal et al, 1998a). Several investigators have overcome this issue by developing different age-appropriate scales (Saigal et al, 1998a).

Although parents are clearly the most knowledgeable source of information about their children, some attributes in a multiattribute health status measure are more difficult to observe than others (Eiser, 1995; Rosenbaum & Saigal, 1996). A parent will know directly, for example, if a child needs help with mobility, but their judgements about emotion or pain are based on the interpretation of their child's behaviour, and thus provide indirect

accounts of HRQoL (Rosenbaum & Saigal, 1996). The ability of parents to rate their child's functioning may also be dependent on demographic factors such as the child's age, sex or health status (Eiser & Morse, 2001a); the accuracy of parental reports has been questioned (Vivier et al, 1994).

Parents' views on their child's HRQoL will be based on different information, different expectations and reflect different experiences than the child or clinician perceives (Eiser & Morse, 2001a). Parental views will also be affected by their own mental and physical health, burden of care giving, wellbeing, concerns, and perceptions of the consequences of disease (Eiser & Morse, 2001a). Parental anxiety and adjustment levels may also affect parental reports (Levi & Drotar, 1998). Eiser and Morse (2001a) found limited support for the widely held view that parents are more able to judge their child's HRQoL in terms of physical rather than social or emotional dimensions (Eiser & Morse, 2001a).

Parents are commonly used as the major informants in child HRQoL assessments, partly owing to the questionable validity of children's self-reports of their HRQoL (Levi & Drotar, 1998). Children have a tendency toward position bias (e.g. tendency to choose the first answer), acquiescence response bias (tendency to agree with researchers regardless of the content of the question), positive response bias (tendency to say 'yes' to questions), limited understanding of negatively worded items, and time perception differences (Pantell & Lewis, 1987; Lawford, Volavka & Eiser, 2001). Children may be too young or too ill to respond, and parents are believed to be the most familiar with their child's life (Rosenbaum & Saigal, 1996).

There is a limit to what can be ascertained first hand, particularly for young children, but this does not mean nothing can be done (Starfield, 1987). The ability of children to complete their own HRQoL assessment partly relies upon the capacity of children to comprehend the cognitive tasks involved in the assessment of HRQoL, and the purpose of the inquiry (Stein & Jessop, 1990; Bradlyn et al, 1993; Torrance et al, 1995; Rosenbaum & Saigal, 1996; Levi & Drotar, 1998; Eiser & Morse, 2001a). Several child-complete HRQoL measures have been developed (Chapter 2), but the minimum age for self-completion is generally five years.

Few studies have systematically investigated whether there is concordance between information regarding child health and child HRQoL obtained from children and other

respondents such as parents, teachers or health professionals (Saigal et al, 1998a). Concordance between parent and child ratings, however, may be expected to increase with the child's age, particularly as greater verbal skills may facilitate children's abilities to describe their experiences and emotions to parents (Eiser & Morse, 2001a). Parent-child concordance in HRQoL outcomes was rarely examined for children less than seven years of age (Eiser & Morse, 2001a). Agreement between child and parent HRQoL scores has been shown to relate to background variables such as sex, age, temporary illness, age of parents and visits to a physician (Theunissen et al, 1998). Eiser & Morse (2001b) found that agreement in the rating of HRQoL is better between parents and chronically sick children compared with parents and their healthy children. They also state that where children report a QoL very different from that reported by their proxy, there is no way of knowing whether the views of the child or proxy reflect the 'truer' picture (Eiser & Morse, 2001a).

A multi-informant assessment of HRQoL, including children's own reports, is therefore recommended as parents are not with their child in every setting in which they function. Medical staff are useful sources of information regarding the HRQoL of children, as they are familiar with symptoms and functional limitations associated with childhood diseases and illnesses (Levi & Drotar, 1998). However, they do not have first hand exposure to the child's life in school, relationship with peers, and feelings about their own health condition (Eiser, 1995). Parent-health care professional differences in judgements of children's HRQoL have also been observed (Cadman, Goldsmith & Bashim, 1984; Pantell & Lewis, 1987; Barr et al, 1994; Rosenbaum & Saigal, 1996; Manificat et al, 1999).

A final barrier reported to the development and adoption of HRQoL measures for children is economic, with expenditure on child health care being much less than that devoted to the health care of adults (Schor, 1998). Therefore, little support for research on child health or child health services is available, resulting in minimal available expertise in the field of HRQoL assessment in children (Schor, 1998).

Pal (1996) recommends that practical solutions to the administration of QoL measures to children of differing developmental levels, and of adjusting definitions of QoL for those levels, need to be devised; healthy children also need to be studied to define the range of physical, psychological and social experiences that might be expected to occur and how

these are affected over a course of time. Pal (1996) suggests that this would aid the interpretation of results from ill children and guide efforts at intervention.

1.2 Paediatric intensive care (PIC)

Paediatric intensive care delivers care to the most critically ill children, most of whom will be artificially ventilated (NHSE, 1997). It is a low volume, high cost service; requires highly trained specialist staff; and cannot be provided in every locality but should be available to all children who need it regardless of where they live (NHSE, 1997). Apart from the newborn that usually undergo treatment in a neonatal intensive care unit (NICU), children who become critically ill at any time between early infancy and adolescence (usually up to 16 years of age) require the facilities of a PICU (Paediatric Intensive Care Society – PICS, 1996). A critically ill child may need more than one of three or four types of care during a single period of treatment in hospital (Table 1.1). Paediatric intensive care is an expensive service, accounting for 20% of the hospital bill (Heaf, 1986) and more recently £950-£1200 per single bed-day for a Level 2 child (NHSE, 1997).

Level of care	Nurse: Patient Ratio	Description
High dependency (Level 1)	0.5: 1	• Requirement for closer observation & monitoring than is available on a standard children's ward • Single organ support, excluding respiratory support • Step down from intensive care (ICU) • Following major surgery: cardiac, neuro, spinal etc. • Advanced analgesic techniques • Non-intubated children with moderately severe croup, bronchiolitis, etc. • The recently extubated child • Children requiring long-term chronic ventilation (with tracheostomy)
Intensive Care (Level 2)	1:1	• Requirement of continuous nursing supervision • Advanced respiratory support Or • Two or more organs systems requiring support Or • One acute organ failure receiving support, plus one chronic failure • Unstable non-intubated child
Intensive Care (Level 3)	1.5:1 and 2:1	• Requirement of intensive supervision at all times • Two or more organ systems requiring technological support, including advanced respiratory support as one of these systems, e.g. renal support or haemofiltration
Intensive Care (Level 4)	2:1 or more	• Child requiring the most intensive interventions, e.g. a level 3 child managed in a cubicle, those on ECMO (extra-corporeal membrane oxygenation), and children requiring renal replacement therapy

*Adapted from '*Standards for PIC*' (PICS, 1996) and '*Framework for the Future*' report (NHSE, 1997).

Table 1.1 Levels of PIC*

1.2.1 Demand for PIC and epidemiology

In March 1997, 280 dedicated intensive care, and specialist intensive care and high dependency beds were identified for children in England (NHSE, 1997). At this time, the Northern and Yorkshire Region had the second highest concentration of intensive care and high dependency beds (n=47), with North Thames having the most beds (n=59). The annual intensive care admission rate for residents of the former Yorkshire Region up to and including 16 years was 1.05 per 1000 for 1995-1996 and 0.99 per 1000 for 1996-1997 (Fairfield, 1997). During 1995-1996 there were a greater proportion of Level 2 and 3 children (93%) receiving intensive care than in 1996-1997 (85%) (Fairfield, 1997).

A wide range of conditions can cause children to become critically ill: from common childhood illnesses such as croup, bronchiolitis or gastroenteritis, to congenital malformations, which increase the risk of critical illness and reduces the best outcome, or inborn errors of metabolism, or acquired conditions such as trauma, seizures or septic shock (Segedin, 1999). Factors such as family income, standard of housing, and parental education strongly influence the risk of a child becoming critically ill (Segedin, 1999). Some critical childhood illnesses, e.g. sickle cell anaemia and cystic fibrosis, are more common in specific racial groups.

There is a higher incidence of severe respiratory illness in autumn and early winter, which substantially increases presentation with critical illness during those months (Segedin, 1999). Between one-quarter and one-third of all children requiring PICU admission in a developed country have a disease of the upper or lower respiratory tract. Most cases are due to viral infections causing croup, bronchiolitis, pneumonia or precipitating acute asthma. Bacterial infections such as pneumonia, whooping cough, epiglottitis or retropharyngeal abscess are less common but carry significant morbidity (Segedin, 1999). Most severe respiratory illness occurs in the first three years of life; bronchiolitis and pertussis in the first six months and pneumonia in the first two years are responsible for much of the mortality and morbidity (Segedin, 1999). Acute respiratory infections account for 50% of all illnesses in children under five years old and 33% of all illnesses in those aged 5-12 years (Barry & Hocking, 1995; Segedin, 1999). It has been reported that 40% of PICU admissions relate to congenital heart disease, 20% to respiratory disease, 15% to major trauma, and less than 10% to neurological problems, the remainder varied depending

upon the allocation of neonatal surgical patients and other services (PICS, 2001). The majority (70%) of UK PICU admissions are unplanned (PICS, 2001).

Most critically ill children remain so for only a short time (1-4 days) during an acute acquired illness or following major surgery. The average length of stay in intensive care units (ICUs) in the Yorkshire region ranged from 4.5 days (1995-1996) to 4.8 days (1996-1997) (Fairfield, 1997). About 1.2% of children in developed countries will become sufficiently ill to require ICU admission. Up to 10% of these children are less than one month old, more than 50% are less than two years old and two-thirds are less than five years old (Segedin, 1999). It has been reported previously that 62% of all PICU admissions in the UK are children aged 0-4 years (BPA, 1993). Over half the children requiring intensive care in Yorkshire (1995/96) were less than two years of age (Fairfield, 1997; NHSE, 1997); this is consistent with national data (Barry & Hocking, 1995; PICS, 2001).

1.2.2 The need for measures of effectiveness of PIC

Paediatric intensive care is a relatively young specialty, which developed in the 1950's and 1960's alongside the specialties of neonatalogy and newborn intensive care, paediatric general surgery, paediatric cardiac surgery, adult respiratory intensive care and paediatric anaesthesiology (Sinclair, 1997). The first ICUs opened after improved outcomes were realised when specialised care was provided to critically ill neonates and adults (Curley, 1996). The establishment of the early PICU's and their associated training programs had an apparently favourable impact on mortality and morbidity outcomes, especially those associated with acute respiratory failure (Downes, 1992). However, the difficult task of following surviving patients to determine long-term outcome was undertaken only by a few centres in the 1980's (Bray & Morrell, 1982).

Despite the relatively young age of PICUs, the development of PIC medicine and new technologies has expanded dramatically, yet evidence describing their clinical and cost-effectiveness is limited. Methodological difficulties have been encountered when conducting research in this field, which may contribute to the lack of studies evaluating the effectiveness of PIC. The choice of suitable outcome measures to evaluate the effectiveness of PIC has also focused on mortality outcomes alone; other outcome measures such as those that evaluate the quality of survival for children discharged from PIC have not been included routinely.

In recent years there has been an increase in the number of children with congenital, previously lethal defects, now being submitted to temporised and prolonged high technology care (Gemke & Bonsel, 1996). Although mortality rates have consequently reduced, the quality of survival in these children remains uncertain (Gemke & Bonsel, 1996). The need for more information about the outcomes for children, who are cared for in different clinical settings related to the severity of illness when they enter the PICU, was also recognised as a priority (NHSE, 1996b). The availability and organisation of intensive care services for critically ill children also needed improving (NHSE, 1996c). The need for outcome measurement in PIC was also reiterated in the *'Framework for the Future'* report that stated that detailed evaluation of PIC, including the collection of data on outcomes, was unsatisfactory (NHSE, 1997). The NHS Research and Development Executive also identified outcomes of PIC as a national priority area for research (NHSE, 1996d). The National Co-ordinating Group (authors of the NHSE report, 1997) believe that other outcome measures should be developed which reflect QoL – including morbidity, disability and functional health of the child after a period in intensive care. They state that in the longer-term the measurement of the humanity and equity of PIC needs consideration (NHSE, 1997). There is currently insufficient evidence available in the UK to inform health care decisions (Fulbrook & Foxcroft, 1999). Although a variety of measures are available that can be used to evaluate child HRQoL outcomes, very few have been reported as valid and reliable for use in a PIC setting.

Most studies investigating cost-effectiveness of intensive care are in the context of neonatal intensive care (NIC) (Kenworthy, Bess & Stahiman, 1987; Parry, 1995; Hall et al, 1995; Stevenson et al, 1996a & 1996b) or adult intensive care (AIC) (Edbrooke et al, 1995; Howard et al, 1995; Stevens, Hibbert & Edbrooke, 1998). Limited studies report the costing of PIC (UK Collaborative Extra Corporeal Membrane Oxygenation (ECMO) Trial Group, 1996), although several studies report the evaluation of resource utilisation in PIC in relation to severity of illness (Klem et al, 1990; McAloon et al, 1991). The latter studies are cost description studies, and not full economic evaluations of PIC; the total patient care costs were not evaluated and only the patient perspective was considered when ideally the societal perspective is preferred in full economic evaluations (Drummond et al, 1997). A clinical score, the Therapeutic Intervention Scoring System (TISS) score, has been described as the most important determinant to calculate total direct medical costs based on

a limited number of readily available clinical variables related to patient characteristics and treatment in a Dutch PICU (de Keizer, et al, 1998).

1.2.3 Measurement issues in the assessment of PIC effectiveness

The paucity of experimental research studies in PIC, such as RCTs with economic evaluations, suggests that intensive care is so self-evidently beneficial it might be unethical to conduct an RCT in this setting (Black, 1996). For example, it is most unlikely that any ethics committee in an industrialised country would sanction the random allocation of patients to intensive care versus ward care (Black, 1996). Other limitations in conducting RCTs in the PIC environment include achieving an adequate sample size. It may take several years to recruit enough eligible patients to the trial, therefore impacting upon the cost of conducting the trial. Multi-centre trials may overcome the problem of recruitment rates, as illustrated by the UK ECMO trial, but will be complex to co-ordinate as a result (UK Collaborative ECMO Trial Group, 1996). When the outcomes of interest are far in the future, the practical difficulties in maintaining such prolonged prospective studies (whether experimental or observational) are considerable, as are their costs (Black, 1996). This is true of the UK ECMO trial, which aimed to assess the QoL for all trial survivors in four and seven-year follow-up studies. The results of this trial should be treated with caution until evidence about the longer-term effects emerges (Roberts & The ECMO Economics Working Group, 1998). Observational studies, and measures of effectiveness, can provide an alternative to leaving the question of the effectiveness of an expensive service such as intensive care unevaluated (Black, 1996). Observational studies in the PIC setting have been reported in the literature, but are few in number (Singh-Naz et al, 1996).

In addition to methodological difficulties described, other factors relating to the delivery of PIC services and the PIC population itself have been reported to hamper the evaluation of the effectiveness of PIC (Gemke, 1999). For example, the PIC population encompasses diverse patient groups with differing levels of severity and types of illnesses. Children who are admitted to PICU following an acute illness or injury are different from those admitted after major elective surgery (Gemke, 1999). It is also difficult to separate the contribution of pre-PICU treatment from that of PICU treatment. The PIC population is particularly heterogenous with differing treatment options available in non-tertiary and tertiary PICUs; tertiary PICUs may offer specialist services such as cardiac and neurological care (Gemke,

1999). Sometimes children are admitted to multidisciplinary adult ICUs where there is a lack of expertise in caring for critically ill children (Gemke, 1999).

1.3 Measures of effectiveness in PIC

The clinical effectiveness of PIC has been described in evaluative studies using several measures of health outcome (Gemke, 1999). These measures include mortality outcomes, service delivery outcomes, functional outcomes, psychological and/or emotional outcomes, and QoL and HRQoL outcomes.

1.3.1 Mortality as an outcome of PIC

Mortality outcomes are reported as a standard measure of health outcome in PIC, with large variations reported between PICUs; 5-15% in multi-centre studies (Pollack, Cuerdson & Getson, 1993; Barry & Hocking, 1995; Gemke, 1999). Mortality outcomes provide an objective measure of health outcome, like clinical and laboratory measures, thus representing a biomedical model of health and illness (Jenkinson, 1994; Gortmaker et al, 1998). Other objective measures also include the presence or absence of disease, the effect of the disease on an organ, and the consequences of the disease in terms of morbidity, incidence, prevalence, mortality rates, case severity, adverse reactions, complications, symptom relief, pain and cost-effectiveness (Bowling, 1995; Garratt, 1998).

Mortality outcomes are attractive to use because death is perceived as an important health outcome by clinicians, and it is easy to measure (MacKeigan & Pathak, 1992). However, in a society where chronic diseases are becoming increasingly prevalent, the objective of health care treatment is more often a reduction in morbidity (e.g. disease, disability & discomfort) than it is a cure of the disease or prolongation of life (Lohr, 1988; MacKeigan & Pathak, 1992). Mortality and morbidity outcomes have been criticised as being insufficiently sensitive and comprehensive to measure health outcome (Vivier et al, 1994). Although mortality outcomes and other objective measures of outcome, such as physiological variables, can provide relevant information to clinicians, they are of limited value to patients, as they do not offer insights into the subjective aspects of health (Donovan, Frankel & Eyles, 1993). Objective measures of health outcome often correlate poorly with functional capacity and wellbeing, the areas in which patients are most interested and familiar (Guyatt et al, 1993). Mortality data is limited and crude mortality rates are unsuitable for PIC inter-institutional comparisons because they are biased by

case-mix variation (Gemke & Bonsel, 1995; Fulbrook & Foxcroft, 1999). Therefore, to make inferences from observed differences in mortality among patient groups in different units, it is essential to collect data on severity of illness and measure and adjust for those aspects of case-mix which influence prognoses (Intensive Care National Audit and Research Centre - ICNARC, 1995).

Subjective health measures can amplify data obtainable from morbidity and mortality statistics by describing the quality rather than the quantity of survival (Donovan et al, 1993; McDowell & Newell, 1996; Fitzpatrick et al, 1998a). They rely on the judgements of individuals, whether patients or clinicians and cannot be ascertained from laboratory tests or population statistics (Donovan et al, 1993). These subjective, patient-based measures of health outcome can provide supplementary information to traditional biomedical assessments by identifying those outcomes valued by patients (Guyatt et al, 1993; Jenkinson & McGee, 1998). The inherently subjective source of patient-based material leaves grounds for anxiety about robustness and scientific value in the minds of some clinicians (Fitzpatrick et al, 1998a). There are also circumstances where patients are unable to provide their unique report of their perceptions due to ill health, physical or cognitive problems, or some other incapacity; in these cases, proxy reports may be necessary (Fitzpatrick et al, 1998a).

Standardised mortality ratios or SMRs (ratio of observed mortality and expected mortality adjusted for severity of illness) can be used to compare the effectiveness of PIC, or of the whole PICU, with that in a large reference population of similar populations in different PICUs (Gemke, 1999). If the observed mortality is lower than the expected mortality, adjusted for severity of illness (SMR<1), this suggests a superior performance in the study group compared with the reference PICU population. Studies have shown that substantially higher SMRs were found in non-tertiary PICUs where the severity of illness adjusted odds ratio for dying was at least two times higher than in the tertiary PICUs (Gemke, 1999). However, discrepancies between observed and expected mortality rates can occur where the child has an underlying chronic illness, or if there are local differences in practice in withholding and withdrawal of treatment (Gemke, 1999). The concept of relating expected future health status (based on baseline health status), with actual (observed) health status is denoted with the standardised health ratio, which in combination with SMRs may become

a new comprehensive indicator of performance in intensive care medicine (de Keizer et al, 1997).

Although intensive care has probably reduced the acute short-term mortality rate of many illnesses, survival to PICU or hospital discharge is not the only end point of effectiveness of advanced medical care. Decreased mortality may be accompanied by an increasing proportion of patients with chronic residual morbidity, so functional outcome and longitudinal assessments of length and quality of survival have become important supplementary measures of outcome after intensive care (Gemke, 1999). Mortality statistics alone are not sufficiently sensitive to detect many important problems with health, and indices of morbidity are important adjuncts (Fiser, 1992).

1.3.1.1 Severity of illness scores in PIC

Comparison of health outcomes in different PICUs is hampered by differences in severity of illness and in case-mix patterns (Gemke, 1999). Clinical scoring systems have been developed to take account of these differences, by adjusting the mortality rate in a PICU, a city, or a country objectively according to severity of illness, in order to address questions of PIC effectiveness, efficiency, and quality of care (Gemke, 1999). Clinical scores and scales are a different kind of subjective perceptual evidence; they are the perceptual judgement of doctors or other health professionals (Fitzpatrick et al, 1998a). Physiologic measures generally provide information to clinicians but are of limited value to patients.

Three clinical scores are currently available for objective and uniform assessment of severity of illness in PICU patients, which enables comparisons to be made between children of different ages, diagnoses, racial and social backgrounds, and pre-existing illnesses (Gemke, 1999). These scores are PRISM (Paediatric Risk of Mortality) developed in the USA (Pollack, Ruttiman & Getson, 1988a), PIM (Paediatric Index of Mortality) developed in Australia and the UK (Shann et al, 1997), and TISS, developed in the USA (Cullen et al, 1974; Yeh et al, 1982).

Both PRISM and PIM measures compare observed with predicted mortality as a measure of the quality of care delivered by a PICU (Randolph, 1997). The TISS measure differs in that it quantifies severity of illness according to therapeutic interventions received by the patient; it can be used to compare different patient populations (Yeh et al, 1982). Both

PRISM and PIM measures have been shown to be valid by accurately predicting death for patients who die and survival for those who live (Randolph, 1997). However, criticisms concerning the validity of the outcome prediction of PRISM have been based on the time at which the variables are recorded, 12 or 24 hours (Fulbrook & Foxcroft, 1999). Many deaths occur during the first 12-24 hours of PICU admission, therefore PRISM may be diagnosing death rather than predicting it in some children (Shann et al, 1997). Furthermore, 12 or 24-hour scores will be affected by treatment given after admission to PICU, so PRISM is not a valid measure for comparing the quality of care between different units or within a single unit over time. Children admitted to a good PICU who recover will have lower PRISM scores than similar children admitted to a bad PICU who are mismanaged in the first 12-24 hours, and the bad unit's high mortality rate will be incorrectly attributed to its having sicker patients (Shann et al, 1997).

The inter-rater reliability of PIM and PRISM measures is reported as satisfactory (Pollack, Patel & Ruttiman, 1996; Shann et al, 1997), except the performance of PIM drops for non-cardiac post-operative patients (Randolph, 1997). In terms of accuracy of the two scores, PRISM has over predicted mortality in comparison to PIM (Shann et al, 1997). The PIM measure clearly outperforms the PRISM measure in ease of use; data is required for only eight variables with PIM compared to the twenty-three required for PRISM. Automated data collection systems could facilitate data collection for PRISM (Randolph, 1997). The coefficients for PIM are freely available, whereas the use of PRISM requires the payment of an annual licence fee (Shann et al, 1997). The PRISM measure has been revalidated at regular intervals by its developers; the maintenance of PIM is less clear (Randolph, 1997). No consensus has been reached as to which measure constitutes the 'gold standard' (Randolph, 1997). Although severity of illness scores can predict mortality in a PICU, they do not predict outcomes such as length of stay or functional status very well and their cost-effectiveness has not been assessed (Randolph, 1997).

1.3.2 Service delivery as an outcome of PIC

The impact of different service delivery processes on the outcome of PIC in the USA is reported (Pollack et al, 1988b; Pollack et al, 1991). There is currently no research evidence available to assess service delivery outcomes in PIC in the UK (Fulbrook & Foxcroft, 1999). Although the reported studies investigate the impact of differing methods of service delivery on PIC outcome, such as tertiary versus non-tertiary care, and intensivist versus

non-intensivist care, outcomes were not evaluated from the child or parent perspective. The main outcome measures used included severity of illness adjusted mortality rates and the incidence of therapeutic and monitoring modalities. The robustness of these studies can be criticised. The results of the study by Pollack et al (1988a) should be interpreted with extreme caution as there were a small number of patients in the moderate and high risk categories, and wide confidence intervals reported suggest that the logistic regression models employed may not be very stable (Teres & Lieberman, 1991). The results of the study by Pollack et al (1991) are also misleading as the medical cover on the PICU changed with the arrival of the intensivist; the broader organisational changes in medical staffing may have resulted in the more favourable outcomes post-intensivist arrival, rather than sole intensivist care. New therapies may also have produced more favourable outcomes.

1.3.3 Functional outcomes of PIC

The effectiveness of PIC has been evaluated in terms of measures of functional outcome. Several measures are available which assess physical disability in terms of functional outcomes following serious or critical illness in children, e.g. the Pediatric Evaluation of Disability Inventory – PEDI (Haley et al, 1989; Feldman, Haley & Coryell, 1990), Functional Disability Inventory - FDI (Walker & Greene, 1991), Functional Independence Measure for Children – Wee-FIM (Msall et al, 1993), and the Klein-Bell Activities of Daily Living (ADL) Schedule for Children (Law & Usher, 1998). Function, within a disability framework, is the ability of a child to perform daily activities independently and safely within the environment (Haley, Coster & Ludlow, 1991). Functional outcomes emphasise performance of activities and are less concerned with the form of the behaviour, i.e. independence rather than normal function (Haley et al, 1991). Measures of functional status in children focus on the child's functional ability to perform age-appropriate daily activities (self-care, mobility, physical activity, role activities – play, and leisure activities), and their overt behaviour, as opposed to their subjective experience with a condition (Speith & Harris, 1996; Levi & Drotar, 1998). Functional status is the objective degree of impairment caused by an illness, whereas QoL also includes the subjective evaluation of the impairment (Speith & Harris, 1996).

Several studies report the evaluation of functional outcomes for children following PIC in the UK, USA and Australia (Bray & Morrell, 1982; Pollack, Wilkinson & Glass, 1987;

Butt et al, 1990; Fiser, 1992; Madagame et al, 1995; Mok & Butt, 1996). There is no consistency in the measures selected to evaluate functional outcomes in these studies. The number of dimensions of health described within the chosen functional outcome measures also varies, ranging from two to three dimensions. Bray and Morrell (1982) reported functional outcomes in terms of physical, behavioural and cognitive outcomes using non-standardised methods. Pollack et al (1987) reported functional outcomes in terms of neurological function and level of independence; they developed their own classification system. Level of independence or level of handicap, and cognitive and motor neurologic function were reported by Butt et al (1990) and Mok and Butt (1996), but the definitions of handicap were not clearly stated. Fiser (1992) evaluated functional outcome in terms of physical and cognitive disability using two scales developed for this purpose, the Pediatric Overall Performance Category (POPC) and the Pediatric Cerebral Performance Category (PCPC). These scales were adapted for use in children from adult scales describing a degree of brain damage. They have been criticised for their imprecision and inter-rater reliability (Gemke, van Vught & Bonsel, 1993). The POPC and PCPC scales also evaluate functional outcome in only two dimensions of health, a limitation acknowledged by the scale developer.

All of the studies reported measure functional outcome in children following PIC from the clinician perspective; parents were not always involved in the assessment. A variety of methods were also employed in the clinician assessments including medical record review; medical or nurse assessments, sometimes using validated measures; and questionnaire or telephone contact with the child's clinician, general practitioner (GP) or parents. The studies also varied in their level of rigour with some utilising different methods of data collection to assess outcome (Butt et al, 1990). Others used a measure developed in a different population to the one in which it was intended (Madagame et al, 1995). Outcomes were often evaluated using only one assessment, so the long-term effects of PIC were not evaluated (Bray & Morrell, 1982). Although these studies build upon those describing mortality outcomes, functional outcomes were only assessed in a few dimensions of health, most commonly physical and cognitive functioning. The views of children were not represented and few parents participated; children and parents might have valued functional outcomes differently to clinicians.

1.3.4 Psychological and emotional outcomes of PIC

Psychological and emotional outcomes have also been measured to evaluate the effectiveness of PIC. A significant proportion of children suffer emotional and behavioural disturbances following hospitalisation, which are exacerbated by prolonged or repeated admissions (Bonn, 1994). Some children discharged from the PICU may experience post-traumatic stress disorder (PTSD) including symptoms of distressful recollections of the traumatic event, avoidance of stimuli associated with the trauma, a range of signs of increased physiological arousal, extreme fear of separation from parents, anxiety triggered by PICU-like incidents (beeping alarms), persistent night terrors, and regressive or infantile behaviour (Gemke, 1999).

Recent research by Rennick & Johnston (2000) has attempted to adapt an adult PTSD measure for use with children – The Children's Impact of Events Scale (Horowitz, Wilner & Alvarez, 1979). This scale has been piloted in fourteen children aged 7-14 years, but further research is needed to assess its reliability and validity. A Children's PTSD Reaction Index is available (Frederick & Pynoos, 1988); it is unclear why Rennick and colleagues did not use this scale in their study. The Pediatric Symptom Checklist is also reported to assess psychological distress in children (Jellineck & Murphy, 1990).

There are few measures available to evaluate the psychological effects of a PICU admission on children and parents, or psychological and emotional outcomes following PIC. Studies describing the psychological and emotional effects of PIC have utilised mainly qualitative approaches, interviewing parents or children about their PICU experience (Scothern et al, 1992; Kendrick, 2000; Noyes, 2000; Playfor et al, 2000; Rennick et al, 2000). Although small-scale, these qualitative studies have provided rich data on children's experiences of PIC. They are valuable as they investigate an under-researched area, children's perceptions of their PICU experience. The involvement of children and their views regarding health and QoL is strongly advocated in the field of QoL research in children (Bullinger & Ravens-Sieberer, 1995; Eiser, 1997; Titman et al, 1997). The aforementioned studies may identify suitable methodologies for eliciting the views of children in a PIC setting, which may aid the future development of HRQoL measures in PIC, including those appropriate for children who are ventilator-dependent. These studies however, have not utilised measures of psychological or emotional outcome. A UK study is currently investigating the psychological sequelae which children

experience following physical recovery from meningococcal disease using a variety of psychological outcome measures (National Research Register (NRR) Project: N0174004460; investigator Professor Elena Garralda). The published findings of this study are awaited. Although the evaluation of psychological or emotional outcome following PIC represents an important aspect of health outcome, it only represents one or two dimensions of health.

1.3.5 HRQoL outcomes of PIC

The evaluation of HRQoL outcomes following PIC is limited to two studies conducted in The Netherlands and Australia (Gemke, Bonsel & van Vught, 1995 & Gemke & Bonsel, 1996; Morrison et al, 2000). These studies utilised differing generic child HRQoL measures, the Health Utilities Index or HUI (Cadman et al, 1986), and the Royal Alexander Hospital for Children Measure of Function or RAHC MOF (Dosseter, Liddle & Mellis, 1996) with the GOS Quality of Life Questionnaire (Graham, Stevenson & Flynn, 1997) (see Chapter 2 for a review of the HUI measure and GOS Quality of Life Questionnaire). The HUI and RAHC MOF are not ideal measures for evaluating HRQoL outcomes in children under the age of five years.

The RAHC MOF is not a multidimensional measure; physical functioning is excluded (Titman et al, 1997). The measure was also developed from the Children's Global Assessment Scale (CGAS), which was used in child psychiatry to describe an index of severity of disturbance rather than HRQoL per se (Shaffer et al, 1983; Titman et al, 1997). The applicable age-range is unclear and there is limited published information on the psychometric properties of the RAH MOF measure. Inter-rater reliability and concurrent validity are reported as moderately good, but further testing of criterion and construct validity is required (Dosseter et al, 1996). The measure may also not be sensitive enough to detect small but clinically important changes in high throughput, less serious conditions (Dosseter et al, 1996). Test-retest reliability of the measure is not reported, but practicality is reported favourably, taking only a few seconds to complete in clinical practice. However, children under the age of ten years had difficulty in completing the measure, and some parents of infants were unable to use the MOF owing to the language used in some of the descriptions, e.g. referring to school when the child was pre-school age (Dosseter et al, 1996). The ability of a global rating scale to provide the same detail as a multidimensional measure is also highly suspect.

More published information is available on the HUI measure (Chapter 2), including the assessment of psychometric properties. The measure is available in English, but was translated into Dutch in this study using published guidelines (Gemke & Bonsel, 1996). The content validity of the HUI was assumed to be good based upon the work of the HUI developers who developed the dimensions of health from the preferences of a random selection of Canadian parents of school children (Cadman et al, 1986). However, the health views of parents from Ontario, Canada, may be culturally different from the health views of parents from The Netherlands. Gemke & Bonsel (1996) believe that the HUI contains universal dimensions and that the strictly functional approach qualifies the measure to be insensitive to differences between North Atlantic countries. The HUI was also developed from interviews with children of age grade seven or eight, older than the population described in this research programme. Therefore, interviews with younger children, or parents of younger children, may have elicited differing dimensions of health or values placed upon them. Further evidence is needed to test the construct validity of the HUI measure using other clinical groups with specific patterns of impairment (Gemke & Bonsel, 1996). Test-retest reliability was not established because the short median length of stay (two days) would require too short an interval between parental interviews to exclude memory affects (Gemke & Bonsel, 1996). Practicality of the HUI measure is described favourably, but completion times were not reported.

The robustness of the findings from these studies can be criticised in terms of the rigour of the research methods employed. Different data collection methods, interviews and questionnaires, were utilised by Gemke et al (1995) and Gemke and Bonsel (1996) for assessing parental perception of a child's health status, which is questionable in a longitudinal study. They also utilised a prospective survey approach, assessing health status in a cohort of children at different points in time (Sapsford & Abbott, 1992; Moser & Kalton, 1996). Such longitudinal designs are appropriate for describing trends in behaviours and attitudes over time (Edwards & Talbot, 1994; Moser & Kalton, 1996), but potential problems may include the retention of subjects (dropout), recall and report responses, and the 'Hawthorne effect' - sample participants may become untypical (Edwards & Talbot, 1994; Moser & Kalton, 1996).

Prospective assessment of pre-admission health status seemed justified as many children were admitted unexpectedly. However, over 50% of the children assessed pre-admission

were admitted for surgery. Therefore, some of these admissions may have been planned and these children perhaps targeted elsewhere prior to ICU admission. Evidence suggests that parents have a decreased ability to think clearly and to problem solve and reduced ability to utilise incoming information when their child is admitted to PICU (Rennick 1986; Hazinski, 1992). Despite this, no problems were reported in interviewing Dutch parents prospectively within 48 hours of PICU admission. Morrison et al (2000) used a retrospective approach and contacted parents 3-24 months following their child's PICU discharge. Retrospective data collection may be prone to recall bias or memory distortions, but has the advantage of speed with results being available as soon as data are collected (Moser & Kalton, 1996). The longitudinal design utilised in both studies may have minimised the bias of seasonal variation.

Both studies utilised a non-random sample drawn from a population of children admitted to one PICU, at a certain point in time. This sample could be interpreted as a cluster sample, i.e. a complete group of units, a probability sample (Moser & Kalton, 1996). Cluster sampling can be used when the sampling frame is unknown and to reduce the field costs of research, but the price to pay is a lack of precision; it is very easy to bias the sample by the choice of one extreme cluster (Sapsford & Abbott, 1992; Robson, 1995; Blacktop, 1996; Moser & Kalton, 1996). Selecting a random cluster of PICU's in both studies may have minimised this bias and provided a more representative sample. The results of both studies cannot be generalised, external validity is poor (Polgar & Thomas, 1995). Gemke et al (1995) and Gemke and Bonsel (1996) post-stratified their sample for referring clinical speciality; other pre-stratifying variables may have been considered to provide a more representative sample of the PIC population in which to generalise results (Fowler, 1993; Moser & Kalton, 1996). Published information was not available on the sampling techniques or inclusion criteria utilised by Morrison et al (2000).

A large proportion of children admitted to PICU are under the age of one year. Gemke & Bonsel (1996) describe the assessment of multidimensional health status in children less than one year of age being hampered by the lack of functional differentiation and the uncertain boundaries of normal. They state that health status in these young children is therefore preferably determined by the testing of neurological development, for which other tests are available, such as the Denver Developmental Screening Test and the Bayley Scales of Infant Development (Gemke & Bonsel, 1996). However, it could be argued that

if an HRQoL measure applicable to children under the age of one year is used as an evaluative measure to detect changes in HRQoL scores for a particular child over time, then the ability of a measure to detect changes in HRQoL scores between different child populations is less important.

Both these descriptive studies have contributed to the advancement of knowledge in HRQoL outcome measurement for children receiving PIC, with the study by Gemke et al (1995) and Gemke and Bonsel (1996) making the most significant contribution using a multidimensional approach. However, both measures employed are not without their own limitations, including age-appropriateness and the language used.

1.4 Thesis aim and objectives

The current literature indicates that there is no suitable measure to assess HRQoL outcomes in children under the age of five years admitted to a PICU with a respiratory illness. This thesis describes a research programme that aims to develop an HRQoL measure for use in this PIC population. The objectives of the thesis are:

- To identify items for inclusion within the HRQoL measure from the literature and theory (Chapter 2)
- To identify the main HRQoL issues of parents (Chapter 3) and clinicians (Chapter 4)
- To develop an HRQoL measure for use in PIC (Chapter 5)
- To establish the reliability, validity and practicality of the HRQoL measure in a PIC population (Chapter 6)
- To discuss the main findings and propose recommendations for future research (Chapter 7).

CHAPTER 2 – Systematic review of child HRQoL measures

2.0 Introduction

This chapter describes a systematic review of child HRQoL measures including generic and respiratory-specific (disease-specific) measures. Guidelines recommended by the NHS Centre for Reviews and Dissemination (CRD) (1996) informed the review process. The current state of knowledge in the field of HRQoL measurement in paediatrics suggests that there is a lack of clarity as to which components belong to HRQoL, and the quality of available measures is uncertain.

The systematic review aims to integrate the evidence on child generic and respiratory-specific HRQoL measures, and to integrate the methods and theories of their development. The systematic review will therefore inform the development of a respiratory-specific HRQoL measure for use in young children following PICU discharge, by identifying those measures with good psychometric properties, and identifying a suitable 'gold standard' HRQoL measure that can be used within the research programme to assess criterion validity of the newly developed HRQoL measure. Several paediatric HRQoL measures have previously been described, but there is a lack of valid and reliable outcome measures for use in a PICU setting (Chapter 1). This review will contribute to the knowledge of HRQoL measurement in young children in a PICU setting.

2.1 Background

Systematic reviews locate, appraise and synthesise evidence from scientific studies in order to provide informative empirical answers to scientific research questions; they are an invaluable first step before carrying out new primary research (NHS CRD, 1996). Three types of systematic review are generally described: statistically integrated or quantitative reviews (meta-analyses), qualitative reviews and methodological reviews (Greenhalgh, 1997a).

Many systematic reviews in health care are undertaken to evaluate the effectiveness of a treatment, clinical intervention or therapy, often using the model described by the Cochrane Collaboration (Edwards, Lilford & Kiauka, 1998; Hutton & Ashcroft, 1998). In these reviews, numerical data are brought together and assessed against methodological (quality) criteria (Edwards et al, 1998). These reviews usually require the analysis of

quantitative data, often using meta-analytic techniques that combine several sources of evidence, which may increase the generalisability of the results and allow a full exploration of the effects in subgroups (Sutton et al, 1998). Methodological reviews differ in that there is no set way to integrate methods. The choice between rival methods relies heavily on argument and ideas about, for example, how bias might be avoided, what is ethical, or what statistical methods are appropriate (Edwards et al, 1998). The review described in this chapter has elements of both a methodological and qualitative review; measures of effectiveness are reviewed qualitatively including the methods employed in their development.

2.1.2 The need for child HRQoL measures in intensive care

Intensive care is an expensive service and requires a large resource but little is known about its effectiveness in children with limited evidence available about health outcomes for children following PIC. The development of generic and patient-based outcome measures reflects a growing concern with the appropriateness of health care to patients, and a need to compare the costs and benefits of services (Stevens et al, 1998). What matters is how the patient feels, rather than how doctors think they ought to feel on the basis of clinical measurements (Bowling, 1997a). Symptom response or survival rates are no longer enough; and particularly where people are treated for chronic or life-threatening conditions, as in intensive care, the therapy has to be evaluated in terms of whether it is more or less likely to lead to an outcome of a life worth living in social and psychological, as well as physical terms (Bowling, 1997a).

Appropriate outcome measures are needed to evaluate HRQoL outcomes in children. Where it is possible to manage but not cure a disease in childhood, it must be determined how far treatment and disease compromise the child's QoL (Eiser & Morse, 2001a). Informed judgements can therefore be made about whether or not treatment is appropriate and where there is a choice, which might be the best option for the child (Eiser & Morse, 2001a). The assessment of a child's HRQoL has been described as important for their protection when they enter a clinical trial of medical intervention (Grodin & Glantz, 1994). In the absence of comprehensive outcome measures, treatment efficacy may be defined purely by biomedical criteria (Schor, 1998). Thus, an increase in survival may be accompanied by a significant rate of impairment and diminished QoL.

2.1.3 Previous research in child HRQoL outcomes

Since the introduction of the concept of QoL, over 20,000 publications have appeared between 1980-1994 in medical research (Ravens-Sieberer & Bullinger, 1997). However, only 13% (n=3050) of these publications relate to children (Ravens-Sieberer & Bullinger, 1997). Bullinger and Ravens-Sieberer (1995) found only 320 publications related to QoL in children. Of these 320 publications, the largest number were in the areas of oncology and transplantation medicine and those conditions with a high mortality, life threatening dimension, and those in which treatment requires a high cost and care effort. Most studies focused on adolescents (aged 13-18 years), some in children (aged 6-12 years), with very few for younger children. Only a small group of studies assessed HRQoL as a multidimensional concept, most relied upon parents or clinicians assessment of child well being (Bullinger & Ravens-Sieberer, 1995). Under ideal circumstances, HRQoL outcome assessment would not be restricted to a single dimension of health, but would rather be multidimensional in order to capture the full range of manifestations of health and illness (Vivier et al, 1994) (Chapter 1).

Within the field of critical care, a recently published systematic review of outcome measures for adult critical care revealed that it was impossible to make clear recommendations as to which particular measures should be used (Hayes et al, 2000). A review of the Cochrane Database of Systematic Reviews (CDSR) (2000) and the Database of Abstracts of Reviews of Effectiveness (DARE) (2000) revealed no systematic review on outcome measures for PIC.

A methodological systematic review of generic and disease-specific measures designed to measure QoL in children with chronic diseases, either by self-report or proxy-raters is reported (Eiser & Morse, 2001a). The review by Eiser and Morse (2001a) identified nineteen generic and twenty-four disease-specific measures; sixteen measures allowed for completion by children and parent/caregiver; seven allowed for completion by proxy, and seventeen allowed for child completion. Eiser and Morse (2001a) investigated the extent of adult measures used in the evaluation of healthcare interventions in children; the appropriateness of adult measures for use in children; the extent of child self-reports corresponding with assessments made by parents and carers; and the feasibility and reliability of proxy measures of QoL in different disease contexts. The review found that adult measures might fail to tap specific aspects of QoL that are important to a child. There

was some evidence of greater concordance between child and parent for physical functioning compared with social and emotional domains, but greater heterogeneity in the latter measures may contribute to inconsistent results (Eiser & Morse, 2001a). Eiser and Morse (2001a) recommend that new HRQoL measures should:

- Follow established procedures for the development of measures
- Take into account theoretical knowledge of children's understanding of illness, emotion, and ability to complete rating scales
- Include facility for child and proxy report
- Include developmentally sensitive age-appropriate sections
- Include generic core and disease-specific modules (Eiser & Morse, 2001a).

2.1.4 Review aim

The systematic review described in this chapter aims to identify HRQoL measures that can be used in the PICU setting to evaluate HRQoL outcomes in young children (<5 years of age). The overall research question for the review is:

How valid, reliable, responsive and practical are generic and respiratory-specific measures for measuring HRQoL outcomes in young children?

A secondary aim is to identify a criterion HRQoL measure, or 'gold standard' measure, to assess concurrent validity of the newly developed HRQoL measure.

2.1.5 Review objectives

The objectives of the review are:

- To identify generic and respiratory-specific HRQoL measures which have been developed for use in young children less than five years of age
- To describe the dimensions of HRQoL used, informed by theoretical principles
- To assess the extent to which these measures have been validated for patient groups
- To make recommendations regarding 'gold standards' for other measures
- To identify further research needs.

2.2 Review methodology

A steering group, comprising a number of clinical and methodological experts provided guidance on the scope of the review and advice on clinical and methodological issues. Experts in the field of outcome measurement guided the development of search strategies with advice from the health sciences librarian (University of York).

2.3 Design

The review is a methodological systematic review identifying HRQoL measures used in studies with children (Polgar & Thomas, 1998).

2.3.1 Inclusion criteria

Studies were included in the review if they met the following criteria:

- Measures of HRQoL, health status, functional status or wellbeing (discriminative, predictive or evaluative). Health status, functional status and QoL are three concepts often used interchangeably in the literature to refer to the same domain of 'health' (Patrick & Bergner, 1990). This is particularly so in the paediatric HRQoL literature where different domains of HRQoL such as the child's functional status, various domains of child health and psychological status, preferences for various health states are described (Levi & Drotar, 1998).
- Measures that have published psychometric properties (some reliability and/or validity). The requirements for a satisfactory outcome measure include the assessment of reliability, validity, responsiveness and practicality (Streiner & Norman, 1995).
- Generic or respiratory-specific, or proxy measures (batteries). Generic or respiratory-specific measures may be applied to the population under study.
- Measures that are child or proxy completed, or both. Patients themselves should ideally provide information on HRQoL. However, children are sometimes too young or too ill to provide information themselves, or they may be too unreliable (Schor, 1998). In this situation, the child's mother is usually asked to provide information. The review therefore included studies reporting HRQoL outcomes from child or proxy respondent (e.g. parent or health care professional) or both.
- Measures that include the child's perspective. Ideally measures should be developed representing the views of the person completing the measure.

However, it may be difficult for young children to be included in the development of a measure (Chapter 1).

- Children aged 16 years or younger. The age-range was specified above the desired age-range of five years as many child HRQoL measures encompass a broad age-range, e.g. 2-18 years. It was important to include all measures appropriate for children under the age of five years as the HRQoL measure was to be developed in this age group.
- English language papers only. The measure is intended for use in a UK population, so measures published in English were considered. Inclusion of papers published in other languages would have greatly increased the costs of the review. Resources were not available for translation. It is recognised that this may introduce bias by excluding papers published in other languages.

2.3.2 Exclusion criteria

Studies were excluded from the review based upon the following criteria:

- Quality of life only measured by clinical indicators, for example, lung function tests or blood chemistry. Clinical indicators do not represent a multidimensional perspective of HRQoL (Chapter 1).
- Quality of life restricted to demographic or environmental indicators, as this does not represent a multidimensional perspective of HRQoL.
- Single dimension measures, for example, pain scales. Single indicators encompass one dimension and can be used to summarise HRQoL data for that particular dimension. However, as single indicators only cover one dimension, combinations of indicators from single HRQoL dimensions may be a more appropriate and useful approach in many research and clinical applications (Spilker, 1996; Levi & Drotar, 1998). The review excluded studies measuring HRQoL as one dimension, as HRQoL is considered a multidimensional concept.
- Review articles or comments about the measurement of QoL in children or adolescents, as this does not meet the purpose of the review to identify the development and testing of measures themselves.

2.3.3 Search procedure

Search strategies were developed using keywords with guidance from outcome searches described by Brettle et al (1998). An example of a search strategy from the Medline database is displayed (Figure 2.1).

MEDLINE

1 quality of life/
2 ((quality adj3 life) or qol or hrqol or hrql or health-related quality of life or health related quality of life).mp
3 ((quality adj adjusted adj life) or qaly).mp
4 health status/
5 ((health adj status) or (health adj state)).mp
6 (health adj utili$).mp
7 (multi adj attribute).mp
8 (global adj health).mp
9 (health adj profile).mp
10 qwb or (quality adj3 (wellbeing or well-being)).mp
11 or/1-10
12 (measure$ or scale? or index or indices or battery or questionnaire).mp
13 (self adj report) or (self-report).mp
14 ((child$ adj report$) or (child-report)).mp
15 ((adolescen$ adj report) or (adolescent-report)).mp
16 ((parent or mother or father or carer or clinician or doctor) and (report)).mp
17 ((proxy adj report) or (proxy-report)).mp
18 or/13-17
19 (child$ or infant or pediatric? or paediatric? or toddler or adolescen$).mp
20 (chq or child health questionnaire or health utilities index or hui).mp
21 respiratory tract diseases/
22 respiratory disorders.mp
23 respiratory system abnormalities/
24 respiratory tract infections/
25 asthma/
26 cystic fibrosis/
27 bronchiolitis/
28 or/21-27
29 animal/
30 human/
31 29 not (29 and 30)
32 28 not 31
33 (Paediatric Asthma Quality of Life Questionnaire or PAQLQ) or (Childhood Asthma Questionnaire or CAQ). mp
34 (reliab$ or valid$).mp
35 11 and (12 or 18)
36 35 and 19
37 20 or 36
38 37 and 34
39 limit 38 to (English language and yr=1980-2000)
40 36 and 32
41 33 or 40
42 41 and 34
43 limit 42 to (English language and yr=1980-2000)

Notation: mp=mapped to subject heading; $ = truncation symbol; ?=plural

Figure 2.1 Medline (index medicus online) database search strategy (1980-2000)

Search terms were applied in combination with the logical operators specified for each electronic database. Several databases were searched to maximise coverage (NHS CRD, 1996). Searches with high recall may have low precision in that they retrieve a large number of inappropriate papers; these problems were partially minimised through the skilled adaptation of search strategies and careful use of indexing terms such as MeSH headings (Lowe & Barnett, 1994). The search process involved an iterative approach, with each search attempt producing relevant studies and further terms to enhance the strategy.

Searching for articles is a process where the introduction of bias can start; publication bias is one example where research yielding statistically significant, interesting, or 'welcome' results is more likely to be submitted and published, or published quickly (Chalmers, Franck & Reitman, 1990; Sutton et al, 1998). Prescott et al (1998) report that about 10% of completed RCTs remain unpublished whilst many others are only published in conference proceedings, particularly if the studies are small and have shown small, non-significant treatment effects. Attempts were made to minimise publication bias by contacting key workers in the field, hand-searching key journals, scanning reference lists of retrieved papers to identify further studies, and reviewing the grey literature (NHS CRD, 1996).

2.3.4 Electronic databases

Electronic databases were searched to identify suitable papers as they have the advantage of covering the literature in many health care areas (NHS CRD, 1996). The Science Citation Index was used to trace citations of important papers through time, to yield further useful references; DARE, which contains English abstracts of reports from the International Network of Agencies for HTA (INAHTA), was searched to identify reports that may contain relevant original data, or cite studies of interest (NHS CRD, 1996).

Several disadvantages are reported in searching electronic databases alone. Electronic databases do not cover all medical journals; Medline indexes about 3,700 journals out of 16,000 published journals (Dickersin, Scherer & Lefebvre, 1994). If a comprehensive systematic review of RCTs depended solely on a Medline search, then about half of the available studies would be omitted (Dickersin et al, 1994). Medline is also a highly structured database with complex indexing rules, which requires a certain level of skill and experience to achieve good (sensitive and specific) results. Medline provides wide coverage of many English language journals and therefore studies published in other

languages may be omitted. Searching EMBASE, which covers many papers in European languages, may minimise this bias, however, non-English papers were excluded from the EMBASE and other searches owing to translation costs.

The following electronic databases were searched from 1980-2000 to increase the possible number of journals:

- MEDLINE (Index Medicus on line) via OVID. Compiled by the National Library of Medicine of the US, indexing about three million medical papers from 1966 (Greenhalgh, 1997b).
- CINAHL (Cumulative Index of Nursing and Allied Health Literature) via OVID, which covers all aspects of nursing, health education, occupational therapy, social services in health care, and other related disciplines from 1983. It indexes 650 English language nursing and allied health journals, books and book chapters (Colclough, 2000a; Greenhalgh, 1997b).
- EMBASE (Excerpta Medica online) via OVID, which focuses on drugs and pharmacology but also includes other biomedical specialities, and is more up to date than Medline and has better European coverage. It indexes more than 3,600 journals and a small number of reports (Colclough, 2000b; Greenhalgh, 1997b).
- PsychLit (now called PsycINFO), which is produced by the American Psychological Association as the computer searchable version of Psychological Abstracts, and covers psychology, psychiatry, and related subjects. Journals are included from 1974 and books from 1987 in English language only (Greenhalgh, 1997b).
- ISI (Institute of Scientific Information) Science Citation Index & Social Sciences Citation Index via BIDS (Bath Information and Data Services) (now available via ISI Web of Science), which indexes references cited in papers as well as the usual author, title, abstract, and citation of papers themselves. It is useful for finding follow up work done on a key paper and for tracking down addresses of authors, and is available from 1981 (Greenhalgh, 1997b).
- Cochrane Library, The Cochrane Controlled Trials Register (CCTR), CDSR, DARE, which are updated quarterly (Greenhalgh, 1997b).

- National Research Register (NRR), which indexes all NHS-funded research projects.
- HMIC (Health Management Information Consortium) via ARC (University of York), which provides access to three bibliographic databases (Dhdata - UK Department of Health Library and Information Service's database; King's Fund Database - holds records of the material of the library of the King's Fund, UK, post 1979; and HELMIS - Health Management Information Service database produced by the Nuffield Institute for Health, University of Leeds, UK, post 1983), covering health management and related topics. Over 250,000 records citing journal article, monographs and technical reports are recorded (Colclough, 1998).
- Index to theses, indexing theses accepted for higher degrees in UK universities from 1950 (Colclough, 1999)
- Dissertation Abstracts International (DAI), indexing theses written at almost all American universities (Colclough, 1999).

2.3.5 Hand searches and scanning reference lists

Quality of life journals were hand-searched from first publication date; paediatric and intensive care journals were hand-searched for the previous 10 years (1990-2000), to identify very recent publications that have yet to be cited or entered and indexed on electronic databases (NHS CRD, 1996). The following key journals were hand-searched, as advised by experts:

Quality of life related journals:

- *'Medical Care'* (1963-2000)
- *'Social Science and Medicine'* (1967-2000)
- *'Journal of Chronic Diseases'* (1955-1996), later *Journal of Clinical Epidemiology* (1996-2000)
- *'Quality of Life Research'* (1992-2000)

Paediatric journals:

- *'Archives of Disease in Childhood'* (1990-2000)
- *'Pediatrics'* (1990-2000)

- *'Journal of Pediatric Psychology'* (1990-2000)

Intensive care:

- *'Critical Care Medicine'* (1990-2000)

It became evident from hand searches and the results of electronic searches, that most child HRQoL measures were developed in the 1990's. These searches were cross-referenced with reference lists scanned from identified papers which aimed to identify further studies and broaden retrieval into published formats other than journal papers (NHS CRD, 1996).

2.3.6 Grey Literature

Grey literature includes reports, booklets, conference proceedings, technical reports, discussion papers and other formats not indexed on databases (Cook et al, 1993). Searching the grey literature, including registers of clinical trials, and hand-searches may identify unpublished studies and minimise publication bias; however it is a costly process (Dickersin et al, 1995). The Index of Conference Proceedings (The British Library) was hand searched to identify if a search of SIGLE (System for Information on Grey Literature) database was justified. The SIGLE database was not easily accessible to the author and is costly to search. Conference proceedings were searched via the Index of Scientific and Technical Proceedings via BIDS to identify research in progress. The NHS NRR (2000) was also searched to identify current UK research projects. Attempts were made to acquire reports of relevant studies directly from authors, as abstracts of conference proceedings are a notoriously unreliable source of data (NHS CRD, 1996). Quality of life bibliographies and indexes from the *'Quality of Life Research'* journal were hand-searched (1994-2000). Databases on British and worldwide theses were also searched.

The world-wide-web was also searched using Internet search engines to identify QoL links using keywords. The following links were identified:

- Mapi Research Institute: http://www.mapi-research-inst.com
- McMaster University: http://www.fhs.mcmaster.ca.hug/
- International Society for Quality of Life Research: http://www.isoqol.org
- Centre for Child Health Outcomes (Paediatric Quality of Life Questionnaire-PedsQL): http://www.pedsql.org
- HealthAct (Child Health Questionnaire): http://www.healthact.com/chq/

Information regarding the use and development of child HRQoL measures and outcome measurement in PIC was sought from key workers in the field (Table 2.1).

Expert	UK Institution	Expert	International Institution
Kathy Rowan	ICNARC, London	Jeanne Landgraf	HealthAct, Boston, USA
John Weinman	Guy's Hospital, London	James Varni	Children's Hospital, San Diego, USA
Meriel Jenney	Llandough Hospital, South Glamorgan, Wales	David Feeny	University of Alberta, Edmonton, Canada
Penny Upton	Morriston Hospital, Swansea	Renoiud Gemke	Free University Hospital, Amsterdam, The Netherlands
Chris McCabe	University of Sheffield	Anne Morrison	The Children's Hospital at Westmead, Sydney, Australia
Gareth Parry	University of Sheffield	Janet Rennick	Montréal Children's Hospital, Canada
Mike Silverman	University of Leicester	Debra Fiser	University of Arkansas, Little Rock, USA
Daniel Shears	St Mary's Hospital, London		
Adam Glaser	St James's University Hospital, Leeds		

Table 2.1 Contact with experts in the field of paediatric outcome measurement

2.3.7 Data extraction form

Primary research papers that met the review inclusion criteria were coded using a specially devised data extraction form, which did not mirror the traditional data extraction forms described for reviewing effectiveness studies (RCTs). The data extraction form included information on bibliographic details, description of the setting, study population, study methodology, development of the outcome measure, results including validity and reliability (NHS CRD, 1996). Dimensions of health utilised by the HRQoL scale developers were also identified. The data extraction form provides an instrument for the extraction of necessary data from selected studies, accurately and without introducing bias (NHS CRD, 1996). Version one of the data extraction form was informed by the work of Greenhalgh et al (1998) and later modified (Version 2) in light of the systematic review by Eiser and Morse (2001a); the latter version differed in layout only. Version 2 of the form was piloted and the author coded all relevant papers using this form (Appendix I).

The quality of studies was also assessed within this form using guidelines described by Feinstein (1987) and Greenhalgh et al (1998), to make judgements about the validity and reliability of measures described and to undertake recommendations about the usefulness and appropriateness of measures to include in the research programme. The following

criteria were assessed: reliability, validity, responsiveness, practicality, feasibility, user-centredness and clinical utility. A simple scoring system was developed where 2 = prima facie evidence that criterion met; 1 = criterion partially fulfilled; 0 = failed or did not mention criterion (Russell et al, 1998). The total score was derived from the sum of the seven quality criteria divided by the maximum score possible (i.e. 14). Where more than one study used the same measure, a total score for the measure was calculated by assessing the maximum total score for each quality criteria, combining the scores, and dividing by the total maximum score possible. Scores were further refined based upon four criteria (reliability, validity, responsiveness and practicality) described by Streiner and Norman (1995) as essential for an outcome measure, using the afore-mentioned scoring system.

Ideally to minimise reviewer bias and increase the validity of the decision process, all studies should be independently assessed by more than one reviewer and disagreements resolved by reference to the review protocol (NHS CRD, 1996; Russell et al, 1998). This was beyond the scope of this review for timescale and resource reasons, but a second reviewer (Jane Noyes) commented on the design of the data extraction form and used version two to extract data on all papers identified on one child HRQoL measure.

2.3.8 Procedure

First, abstracts were screened in order to assess the relevance of the papers. Papers for inclusion in the review were selected using the afore-mentioned criteria and data extracted using the data extraction form. Papers that did not meet the inclusion criteria were excluded at this stage. If abstracts were ambiguous the full paper was obtained and a decision made to include or exclude the paper based upon the full text.

Relevant references were downloaded into Reference Manager (Reference Information Systems, version 8.0; 1997), a bibliographic software package, to facilitate the production of reference lists. Studies included in the review were displayed in tables including those excluded studies detailing the reasons for each exclusion (NHS CRD, 1996). Papers included in the review were assessed to avoid including multiple publications based on the same data (NHS CRD, 1996).

2.3.9 Second reviewer

A second independent reviewer assessed all papers identified from the review on the development and testing of one child HRQoL measure. Since the purpose of the review is to identify and judge the psychometric properties of child HRQoL measures, the second reviewer also independently judged the psychometric properties of the HRQoL measure using the data extraction form, including assigning a quality score. Responses on the data extraction form and quality scores were compared for agreement. The consistency of the judgement on the reliability and validity of child HRQoL measures was tested using the data extraction form. The second reviewer also checked the literature for additional sources on the identified HRQoL measure and reviewed the data extraction form.

2.3.10 Data analysis

A qualitative overview summarises the aims of the study, a description of the measure used (purpose and development), the procedure (study design, sample characteristics), and study evaluation (instrument development, analysis and results – validity, reliability, responsiveness, practicality, feasibility, and user-centredness). The qualitative overview includes the methodological rigour, and therefore reliability of the studies, also highlighting differences (NHS CRD, 1996). A quantitative synthesis (meta-analysis) was not appropriate in this case as the purpose of the review was not to provide an overall estimate of effectiveness, but to identify child HRQoL measures and review their psychometric properties. The descriptive data was not quantifiable.

2.4 Results

This section provides results for the following sections: i) included and excluded studies, ii) second review, and iii) all relevant measures. The results of the review have been described previously in a qualitative narrative form (Grange, 2001).

2.4.1 Included and excluded studies

For each electronic database searched a total number of abstracts were identified, duplicates between databases were removed at this stage (Table 2.2). Sixty-three publications met the original review inclusion criteria and data was extracted from these publications. Thirty-eight of these publications were excluded further when a more refined criterion of child age of less than five years was applied, resulting in 25 included publications from database searches. The excluded publications from database searches

(n=38) and excluded papers identified from hand-searches (n=13) represented generic and respiratory-specific HRQoL measures (n=21) appropriate for use in children aged five years or more (Appendix II). A further 26 publications were included in the review from hand-searches and citation searching representing 51 publications in total describing 47 studies.

Electronic database	No. of abstracts	No. of abstracts/papers reviewed	No. of papers included
MEDLINE	293	24	15
CINAHL	117	5	0
EMBASE	246	5	3
PsychLit (PsycINFO)	95	7	2
ISI (Web of Science)	78	20	4
Cochrane – CTR	7	0	0
Cochrane – DSR	134	0	0
DARE	25	0	0
NRR	1	0	0
HELMIC	116	2	1
Index to theses	3	0	0
DAI	260	0	0
Database total	-	63	25
Hand-searching and citation searching	-	-	26
Total	-	-	**51**

Table 2.2 Results of systematic review

2.4.2 Results of second reviewer

Findings revealed that although the data extraction from was quite long, the second reviewer completed the form with relative ease, when evaluating the chosen papers. However, some sections of the form were difficult to complete, as often information was not available on the measure in one paper alone.

The author purposefully chose the PedsQL measure for the second reviewer to evaluate as several papers are published on its development. Five papers were identified from the systematic review on the development of this measure (Varni et al 1998a & 1998b; Varni et al, 1999a; Varni, Seid & Kurtin, 1999b; Varni, Seid & Rode, 1999d). A sixth paper (Seid et al, 1999a) was excluded from this evaluation as it described identical data to that published in another paper (Varni et al, 1999c). The second reviewer assessed these papers

to determine the overall psychometric properties of the PedsQL measure; quality scores assigned by the author and second reviewer were compared. The second reviewer also re-ran the search strategy and identified a further five papers on the PedsQL measure. Two papers were conference abstracts (Lawford, unpublished; Seid, Varni & Seizer, 1999b). The first abstract was unpublished and the second although published could not be traced; both were therefore excluded in the final analysis. The other three papers were excluded from the review: one paper was a review paper and did not meet the review criteria, and two papers were published after the review deadline (Varni, Seid & Kurtin, 1999c; Seid, Varni & Kurtin, 2000; Varni, Seid & Kurtin, 2001).

The second reviewer concluded:

- The PedsQL measure was partially reliable and valid
- Internal consistency reliability was reported for the PCQL and PCQL-32 versions, but insufficient data was reported on the PedsQL 4.0 version to assess reliability and validity
- The PedsQL measure partially met the practicality, clinical utility and feasibility criteria. This was supported by evidence of little missing data in the PCQL-32 and minimal floor effects with no ceiling effects
- The PedsQL measure did not meet the user-centredness criteria using evidence published in the papers
- A quality score of five was rated for the PedsQL measure, which agreed with the quality score rated by the author.

2.4.3 Results of all included measures

The included publications described one child respiratory-specific and twelve generic HRQoL measures. The dimensions of health evaluated in these reviewed measures are summarised (Table 2.3). Of the theoretical dimensions of health identified in the review, all included HRQoL measures considered a physical dimension of health with eleven measures considering this as motor or functional development. Six measures did not include an emotional or social dimension of health; seven measures excluded a cognitive dimension of health and only one measure considered a spiritual dimension of health. Two measures included an environmental dimension of health and five measures included a dimension of overall general health perceptions. None of the included measures reviewed identified all widely accepted dimensions of HRQoL, namely physical, emotional, social,

DIMENSIONS - Characteristics	CAQ	RAND	Ontario CHSI	NHIS	FS (II) R	QWB	QoL Nordic	ITQoL	HUI	AUQEI	PedsQL 4.0	QUALIN	WCHMP
PHYSICAL – Motor / functional development		✓	✓	✓	✓	✓	✓	✓	✓	✓	✓		✓
Senses			✓						✓				
Role activity	✓	✓							✓				
Self-care ability		✓	✓										
Pain									✓			✓	
Symptoms	✓					✓							
Fertility									✓				
EMOTIONAL - General									✓	✓	✓	✓	
Positive			✓										
Negative	✓												
Self-esteem										✓			
SOCIAL - General functioning		✓				✓	✓	✓			✓	✓	
Relationships							✓			✓			
Behaviour													✓
Activities (external)										✓			
Activities (leisure)										✓			
Family life							✓			✓			
Environment												✓	
COGNITIVE – General, mental functioning		✓					✓	✓					
School ability			✓								✓		
Cognition / learning			✓						✓				
SPIRITUAL							✓						
ENVIRONMENT - Risks	✓												
Work							✓						
Economy							✓						
Housing							✓						
GENERAL HEALTH – Perceptions		✓			✓								✓
Disorders & treatment				✓						✓			✓*
HRQoL													✓

Table 2.3 Dimensions of health included in measures identified in the systematic review

* Acute minor illness, acute significant illness, immunisation status, chronic illness status, hospital admission status, accident status

and cognitive (mental) functioning. The psychometric properties of each measure were reviewed and a quality score assigned. The included respiratory-specific and generic HRQoL measures are summarised in tables.

Measure 1: Childhood Asthma Questionnaire (CAQ)

Five papers were identified on the CAQ describing six studies (Table 2.4).

Name of measure	CAQ
References	French, Christie & West (1992 & 1994b); Christie et al, (1993); French, Christie & Sowden (1994a); French, Carroll & Christie (1998)
Place of origin	UK (Australian version later developed)
Purpose of measure	Self-complete measure to quantify children's feelings about their everyday lives and their asthma
Item generation	Literature review, discussions with children (French, Christie & West, 1994)
Respondent	Child (three forms available A, B & C)
Age range	CAQA: 4-7 yrs CAQB: 8-11 yrs CAQC: 12-16 yrs
Number of items	CAQA: 14 CAQB: 22 CAQC: 31
Number of dimensions (type)	CAQA: 1 (distress) CAQB: 2 (active & passive QoL) CAQC: 5 (active QoL, teenage QoL, distress, severity & reactivity)
Rating scale	Smiley faces: 4-point CAQA; 5-point CAQB/C
Scale of studies	1) Pilot study: 10 & 15 children; 242 children – CAQB (French et al, 1994b) 2) Pilot study: 21 parents & children; 34 children – CAQA (French et al, 1994b) 3) Pilot study: 25 adolescents – CAQC (French et al, 1994b) 4) 484 children – CAQB (Christie et al, 1993) 5) 535 children – all versions (French et al, 1994a) 6) 784 children – CAQB/C (French et al, 1998)
Sample composition	1) CAQB: 10 healthy & 15 asthmatic children; 214 non-asthmatics 2) 21 children (mean age 6.5 yrs); 34 children (mean age 5 years 4 months) 3) 25 asthmatic adolescents (mean age 13.5 years) 4) CAQB: 270 asthmatics; 214 non-asthmatics 5) CAQA: 80 asthmatics; 103 non-asthmatics CAQB: 103 asthmatics; 153 non-asthmatics CAQC: 98 asthmatics 6) Asthmatics and non-asthmatics
Reliability	Internal consistency - CAQA (α = 0.56-0.63); CAQB (α = 0.21-0.82); and CAQC (α = 0.50-0.80) (French et al, 1994a); CAQB (α = 0.57-0.84) (Christie et al, 1993). Test-retest reliability coefficient 0.70-0.80 (French et al, 1994a)
Validity	Face validity is good; the measure was developed with children. Construct and clinical validity described with differences between scores of asthmatic and non-asthmatic children (French et al, 1998); intraclass correlations (ICC's) also reported. Criterion validity was not formally assessed, but scores were compared with a UK-version of the CAQ.
Responsiveness	Not reported
Practicality	10-15 minutes to complete for all age groups (French et al, 1994a)
Quality score	7 (reliability = 2; validity, practicality, feasibility, user-centredness and utility = 1; responsiveness = 0)

Table 2.4 Summary review of the CAQ (Measure 1)

The CAQ has been extensively tested in large samples of asthmatic and non-asthmatic children, aged 4-16 years, in the UK and Australia (French et al, 1998). The internal consistency of the CAQ is generally satisfactory, but some alpha coefficients were reported as low; this may have been owing to the difference in the number of items in the CAQ compared to other measures as alpha increases with the number of items (French et al, 1994a). Reliability statistics of CAQC compare closely to those of an adult measure (Asthma Quality of life Questionnaire).

This measure shows good psychometric properties (reliability, validity and practicality) but criterion validity or responsiveness is not reported. The measure is not suitable for children under the age of four years; it also focuses on the evaluation of HRQoL in children with asthma alone. The psychometric properties of the measure would require further testing if it were to be employed in this study in children over the age of four years with respiratory illnesses other than asthma.

Measure 2: RAND Health Insurance Experiments (HIE) I & II

One paper was identified on the RAND measure, and one paper on the RAND General Health Rating Index (GHRI), also described in Measure 5 (Table 2.5).

Both infant and child forms were reported to have good discriminant validity, but this varied across dimensions. Scales to measure functional limitations could not be evaluated for either age group as very few children had severe or mild limitations attributable to health. Internal consistency was on the low side but acceptable; some items pertaining to resistance/susceptibility to illness, current health, and mental health, did not consistently correlate as hypothesised.

The measure does not distinguish among children with differing degrees of impaired functioning; it is therefore not sensitive to assess changes in children with chronic illness over time or to measure the wide range of function of children with chronic disorders without major handicaps (Stein & Jessop, 1990). Test-retest reliability was not assessed owing to measurement resources and respondent burden; responsiveness and practicality are not reported. This measure encompasses the age-range of children in this study, but was not considered for use owing to its weak psychometric properties.

Name of measure	RAND HIE I & II
References	Eisen et al (1979); Scholle et al (1995)* (*also described in Measure 5)
Place of origin	Rand Corporation, California, USA
Language	English
Purpose of measure	To test hypotheses about health care financing & health status
Item generation	Literature based & other questionnaires; the measure focuses on physical, mental and social aspects of health.
Respondent	Proxy - parent
Age range	Infant form: 0-4 years Child form: 5-13 years
Number of items	Infant form - 157; Child form – 122; GHRI – 7
Number of dimensions (type)	Infant: 3 – physical health (item groupings of physical activity, role activity & self-care activity – 5 items), general health perceptions (item groupings of current health, resistance/susceptibility & prior health), and developmental milestones (item grouping of satisfaction with development) Child: 4 - physical health (mobility, physical activity, role activity & self-care activity – 13 items), general health perceptions (as before), substitutes developmental milestones for a mental health index (item groupings of anxiety, depression & positive well being) and social health index (item grouping of social relations) GHRI: ? 3 (current health, previous health & resistance to illness)
Rating scale	1) Not reported; 2) GHRI range 8-35
Scale of studies	1) 2,290 families in 5 of 6 HIS (Health Insurance Study) sites in USA (1975-1977) (Eisen et al, 1979) 2) 608 children in 8 hospitals in USA
Sample composition	1) Not reported; 2) pre-term low-birth weight (LBW) children
Reliability	1) Internal consistency reported with α values >0.5; test-retest reliability not reported
Validity	1) Face, discriminant and construct validity is reported (Eisen et al, 1979) 2) GHRI has poor concurrent and predictive validity (Scholle et al, 1995)
Responsiveness	The measure is reported as possessing sufficient variability to allow detection of potential differences in health status (Eisen et al, 1979)
Practicality	Not reported
Quality score	5 (reliability, validity, feasibility, user-centredness and utility = 1; responsiveness and practicality = 0)

Table 2.5 Summary review of the RAND HEI I & II (Measure 2)

Measure 3: Child Health Status Index (CHSI) for Ontario Children

One paper was identified on the CHSI for Ontario children (Table 2.6).

The psychometric properties of the CHSI for Ontario children are weak; internal consistency was adequate, construct validity, responsiveness and practicality were not assessed. The authors failed to clearly address how the measure is to be used. The measure does not cover the age-range of children in this research programme and relies upon interviewer-administration to both parent and child, which is time-consuming and may require interviewer training.

Name of measure	CHSI for Ontario Children
References	Cadman et al (1986)
Place of origin	Ontario, Canada
Language	English
Purpose of measure	To assess the health status of children in Ontario using a health status index for epidemiological, population health & health policy research
Item generation	Literature based – tested in parents and child interviews (Stage 1 & II)
Respondent type	Proxy – parent; & child
Age range	4-16 years
Number of items	? 15 (Stage 1); 6 (Stage II)
Number of dimensions (type)	15: Stage I - physical activity, mobility, self-care, school performance, play, learning ability, happiness, pain or discomfort, sight, hearing, speech, use of limbs, cause of health problem, age of onset of health problem, & name of disease or disorder 6: Stage II - sensory & communication ability, happiness, self-care ability, pain or discomfort, learning & school ability, & physical activity ability
Rating scale	Stage I: importance of 15 attributes ranked on a 0-100 point 'feeling thermometer' Stage II: utility of 6 attributes ranked using category scaling and time trade-off methods to produce single summary index
Scale of studies	Stage 1: 84 parent/child pairs & further 120 parents Stage II: 64 parent/child pairs from Stage I
Sample composition	Stage I: Random sample of 84 parent and child (grade 7 or 8) same-sex pairs; 120 parents of chronically ill children
Reliability	Internal consistency tested by correlation analysis of rating and ranking scores: Stage I – Guttman coefficient of reproducibility >0.9, and for scalability >0.6; Stage II correlations of 0.5-0.9 reported as less satisfactory. Scaling technique showed considerable random error (unreliability)
Validity	Criterion validity only - assessed by comparing health states assessed by parent/child pairs; statistical differences were found
Responsiveness	Not reported
Practicality	Not reported
Quality score	5 (reliability, validity, feasibility, user-centredness and utility = 1; responsiveness and practicality = 0)

Table 2.6 Summary review of the CHSI for Ontario Children (Measure 3)

Measure 4: National Health Interview Survey (NHIS)

Two papers and one report were identified on the NHIS, which identified the health and functional status of a large sample of children in the USA (Table 2.7).

The survey determined the disease burden resulting from a number of chronic childhood diseases, such as asthma, via the assessment of days missed from school, contact with medical staff and hospital admissions. The functional status of children with or without a chronic childhood illness was also determined. Data was used to calculate prevalence rates for chronic illnesses. Results showed that the health status of children with asthma was lower than the health status for other children, and children with asthma experienced more limitations in activity than healthy children.

Name of measure	NHIS
References	Adams & Hardy (1989); Newacheck & Taylor (1992); Taylor & Newacheck (1992)
Place of origin	USA
Language	English
Purpose of measure	To gather epidemiological data on chronic illness in children in the United States of America.
Item generation	Not reported
Respondent type	Proxy – parent or responsible adult family member
Age range	0-18 years
Number of items	? Core questionnaire; 65 questions in supplementary questionnaire
Number of dimensions (type)	Core questionnaire (perceived health status & functional status – limitation of activity & restriction of activity) Supplementary questionnaire on childhood conditions, e.g. asthma
Rating scale	Core questionnaire – Likert-type response format Supplementary questionnaire – continuous data
Scale of studies	Core questionnaire – 47,485 household representing 122,310 persons, including children Supplementary questionnaire - 17,110 children
Sample composition	Random sample of civilian non-institutionalised population residing in USA
Reliability	Not reported
Validity	Not reported
Responsiveness	Not reported
Practicality	Data collected by household interview
Quality score	2 (feasibility and user-centredness = 1; validity, reliability, practicality, responsiveness and utility =0)

Table 2.7 Summary review of the NHIS (Measure 4)

The psychometric properties of the NHIS are unclear and the process of item generation is not reported. The measure focuses on functional health and disability, not health as a multidimensional concept. The views of children were not elicited in the development of the measure. The survey is interviewer-administered which is time-consuming. The measure was not considered for use in this research programme for these reasons.

Measure 5: Functional Status or FS II (R)

Four papers describing four studies were identified on the FS measure (Table 2.8).

The FS II (R) was developed from an earlier version the FS I which assessed behavioural responses to illness that interfered with normal social role performance in three sites (home, neighbourhood & school) during leisure, work, and rest activities. Behavioural statements were created for four age categories: infants (0-9 months), toddlers (9-23 months), pre-schoolers (2-5 years) and school-age children (>5 years).

Name of measure	FS II (R)
References	Stein & Jessop (1982); Stein & Jessop (1990)*; Stein & Jessop (1991)*; Scholle et al (1995); Kromer et al (2000)
Place of origin	New York, USA
Language	English & Spanish
Purpose of measure	To measure the health status of children, particularly those with chronic, physical disorders, to use as a health status indicator to characterise populations
Item generation	Version 1 (FS I): literature based, interviews with mothers & health care providers, & author's own experiences. Modelled on the Sickness Impact Profile for adults, developed in 1978 Version 2 (FS II I): interviews with mothers & data collection by doctors, developed in 1981
Respondent type	Proxy – parent
Age range	0-16 years
Number of items	Long version – 43; short version – 14
Number of dimensions (type)	FS I – 8 (communication, mobility, mood, energy, play, sleep, eating & toileting patterns) FS II I – The long version produces 2 factors – a General Health factor & an age-specific Development/Functioning factor (Responsiveness ≤ 1 year old; Activity 2-3 year olds; Interpersonal Functioning & Locomotion > 4 years old)
Rating scale	Total score & two factor sub-scores; the short version produces a single index score
Scale of studies	1) a) 140, b) 209 & c) 292 children (Stein & Jessop, 1982) 2) 732 children (Stein & Jessop, 1990). No details on FS I. 3) 608 children (Scholle et al, 1995) 4) 115 children (Kromer et al, 2000)
Sample composition	1) a) & b) chronically ill children; b) 40 children <9 months old, 32 children 9 months – 2 years old, 56 children aged 2-4 years, and 81 children aged 5-10 years; c) 140 chronically ill children & 152 children with minor ailments 2) Children with and without chronic physical conditions 3) Pre-term LBW children from 8 US hospitals 4) 115 Hispanic children with asthma
Reliability	Reliability analysis produced alpha coefficients 0.62-0.83 (Stein & Jessop, 1982). Internal consistency of factor-based scales reported >0.8 (Stein & Jessop, 1990). Test-retest reliability was not assessed because of the probing involved by the interviewer in part 2.
Validity	Discriminant validity tested by examining scores of children with/without medical problems; well children had higher mean scores. (Stein & Jessop, 1990). Correlating scores with established measures of morbidity tested concurrent validity; correlations were consistent across age-ranges and scales. Construct validity was tested by a) correlating scores with a clinical rating of limitations in activity; moderate correlations in the expected direction resulted, & b) differences in scores of children with varying levels of impairment; results revealed differences in the expected direction (Stein & Jessop, 1990). Concurrent validity was tested with the RAND GHRI & showed modest correlations (r=0.33) (Scholle et al, 1995)
Responsiveness	Not reported
Practicality	Interviewer-administered to parent, as a probe is needed in Part 2. Simple, low-cost, can be administered by lay-interviewers.
Quality score	6 (reliability, practicality, user-centredness and utility = 1; validity = 2; and responsiveness = 0)

* An error in journal printing, only Stein and Jessop (1990) reviewed.

Table 2.8 Summary review of the FS II (R) (Measure 5)

The FS II (R) is in two parts: part one asks whether the child performs the specified activity or exhibits a specified behaviour; part two probes those items in part one that reflect poor functioning to determine whether a given functional impairment was due to a health problem. Both versions of the FS II (R) measure show good internal consistency, validity and practicality, but the responsiveness of the measure is unclear. The particular strength of the FS II (R) is the measurement of health status of children with chronic physical conditions who are not disabled. This is relevant to part of the population in this research programme, but it is unclear how to compare scores across age groups, as there are a different number of dimensions across the age ranges. For this reason, the measure was not considered for use in this research programme.

Measure 6: Quality of Well-Being (QWB) Scale

One paper was identified on the QWB measure describing one study (Table 2.9).

Name of measure	QWB Scale
References	Bradlyn et al (1993)
Place of origin	USA
Language	English
Purpose of measure	A preference-based multi-attribute system to measure QoL in adults and children to help guide policy decisions
Item generation	Subcomponent of the General Health Policy Model developed for adults
Respondent type	Proxy – parent
Age range	Infancy to old age; 4 -18 years in this study
Number of items	27 symptom-items plus 3 further items
Number of dimensions (type)	Functional status is assessed in 3 areas (physical functioning, social/role functioning, & mobility)
Rating scale	Visual-analogue measure, that is value-scored producing a total score and subscale scores. Quality adjusted life years (QALYs) may be calculated
Scale of studies	30 children (Bradlyn et al, 1993)
Sample composition	Parents of oncology patients who were being treated or were within two years of having completed treatment were interviewed in presence of child (Bradlyn et al, 1993)
Reliability	Internal consistency reported as adequate for each of the subscales and the total QWB score in paediatric oncology patients (Bradlyn et al, 1993)
Validity	Face & content validity poor; the measure was developed in adults. Criterion validity - tested and reported to distinguish children with cancer with differing levels of treatment toxicity as classified by clinicians; but the correlations reported were low. Not reported in child studies; sensitivity & specificity tested in adult studies
Responsiveness	Not reported in child studies, but reported in adult studies
Practicality	Interviewer-administered; 15 minutes to complete
Quality score	5 (reliability, validity, practicality, user-centredness and utility, responsiveness =1; feasibility =0)

Table 2.9 Summary review of the QWB Scale (Measure 6)

The QWB scale is an adult HRQoL measure that has been used to assess the QoL of children with cancer (Bradlyn et al, 1993). It has also been used in children over the age of five years with cystic fibrosis (Orenstein et al, 1989; Munzenberger et al, 1999). The QWB attributes are not structurally independent; i.e. a person cannot score on the lowest level for physical activity and on the highest level on all the other attributes. The symptom-problem complex attribute also represents the bulk of the overall score. The measure shows poor face and content validity, the preference weights were developed in adults and may not be relevant to young children. Construct validity, test-retest reliability and responsiveness were not tested in children, but are reported in adult studies (Bradlyn et al, 1993). Practicality of the measure is supported. The limited information on the psychometric properties, and its development in an adult population, precludes the use of the measure in this research programme.

Measure 7: QoL in children in Nordic countries

Two papers and studies are reported using this measure (Table 2.10).

Face validity of the measure appears satisfactory as the questions were developed from a theoretical perspective, but the theory employed explored the concept of QoL, not HRQoL per se, for children in Nordic countries. It is unclear how the measure was translated for use in each country. This measure is inappropriate for use in children from the UK who are culturally different from Nordic children, and who speak a different language. The extent to which the dimensions of interest were comprehensively sampled by the items or questions in the measure is uncertain.

The psychometric properties of this measure are not evident. The reliability of the measure is uncertain, but the authors report that the validity and reliability of the measure will improve once a follow-up study of special groups of children with disabilities is used and compared to their results. Both parents and children, who were able, completed the questionnaires together, which may have introduced bias. There are no reports of the responsiveness of the measure or its practicality. This measure was therefore not considered for use in this research programme.

Name of measure	QoL Questionnaire for Children in Nordic Countries
References	Lindström & Köhler (1991); Lindström & Eriksson (1993)
Place of origin	The Nordic School of Public Health, Göteborg, Sweden
Language	Administered in 5 Nordic countries (Denmark, Finland, Iceland, Norway & Sweden), uncertain if translated into native language of each country
Purpose of measure	To measure QoL in children as external, inter-personal and personal conditions in objective and subjective ways
Item generation	Theoretical model of QoL originally to measure mental dimensions of QoL, then broadened to include social & economic spheres, then further developed by adding a global sphere.
Respondent type	Proxy – parent, and child
Age range	2-18 years
Number of items	74 or 75
Number of dimensions (type)	4 life spheres each with three dimensions, 12 in total: i) Global – macro-environment, human rights & policies External ii) External – work, economy & housing iii) Inter-personal – family, intimate relationships & extended/social support iv) Personal – physical, mental & spiritual
Rating scale	Not reported
Scale of studies	1) 15,354 children from 5 Nordic countries (Lindström & Eriksson, 1993) 2) 2,253 children from 5 Nordic countries (Lindström & Köhler, 1991)
Sample composition	1) Five random samples of children were chosen from population registers in each country for the reference sample 2) Children with disabilities including cystic fibrosis (CF), haemophilia, myelomeningocele (MM), osteogenesis imperfecta & visual impairments
Reliability	Not reported, under evaluation
Validity	Not reported, under evaluation
Responsiveness	Not reported, under evaluation
Practicality	Postal questionnaire, completion time
Quality score	5 (validity, practicality, feasibility, user-centredness and utility =1; reliability and responsiveness = 0)

Table 2.10 Summary review of the QoL of children in Nordic countries (Measure 7)

Measure 8: Infant Toddler Quality of Life (ITQoL) Questionnaire

One paper was identified on the ITQoL measure (Table 2.11).

The ITQoL questionnaire is currently being developed by Jeanne Landgraf, one of the CHQ developers (Landgraf & Abetz, 1996). There is virtually no published information on this measure (Landgraf, 1999); much of the information about it was obtained from a meeting with the scale developer in Boston, USA (July, 2000).

The ITQoL measure examines parental reports of infant and toddler, aged two months to four years, physical, mental and social health. The psychometric properties of this measure are unknown. The ITQoL measure covers most of the age-range of children in this research programme and also considers the health and wellbeing of parents. The measure was

considered a possibility for use in the research programme as it covered the age-range of interest, but was excluded because of the lack of published information on its psychometric properties, and because the measure is long (104 items). This may result in a considerable burden to parents when completing two HRQoL measures in the assessment of criterion validity.

Name of measure	ITQoL Questionnaire (ITQoL)
References	(Landgraf, 1999)
Place of origin	Boston, USA
Language	English
Purpose of measure	To assess infant HRQoL to assist neonatologists in identifying families "at risk" upon discharge from the neonatal intensive care unit (NICU)
Item generation	Theoretical - review of the infant child health literature
Respondent type	Proxy –parent
Age range	2 months – 4 years
Number of items	104
Number of dimensions (type)	3 (physical, mental & social health) 8 scales assess the health & wellbeing of the child (83 items); 5 scales assess the health & wellbeing of the parent (21 items)
Rating scale	Likert-type questions as in the CHQ
Scale of studies	USA, Canadian & Australian studies in progress
Sample composition	Not known
Reliability	Not reported, under evaluation
Validity	Not reported, under evaluation
Responsiveness	Not reported, under evaluation
Practicality	Not reported, under evaluation
Quality score	0 (no published information available)

Table 2.11 Summary review of the ITQoL questionnaire (Measure 8)

Measure 9: Health Utilities Index II & III or HUI (multi-attribute health status (MAHS) classification)

Twenty-three papers describing eighteen studies were identified using the HUI measure (Table 2.12).

The measure was first developed for use in evaluating outcomes for very low-birthweight infants on NIC (Boyle et al, 1983; Feeny et al, 1995). Torrance et al (1995) state that the face validity of the HUI system is empirically supported despite the numerous levels on each attribute, as all levels on every attribute appeared at least once in the population health surveys. There were no attribute levels that represented non-existent outcomes.

Name of measure	HUI
References	Feeny et al (1992 & 1993); Barr et al (1994, 1997 & 1999); Billson & Walker (1994); Saigal et al (1994a, 1994b, 1998a); Boyle et al (1995); Torrance et al (1995 & 1996); Gemke et al (1995); Gemke & Bonsel (1996); Glaser et al (1997a & 1997b) Trudel et al (1998); Glaser et al (1999); Le Gales et al (1999); Speechley et al (1999)*; Szecket et al (1999); Mulhern (1999); Felder-Puig et al (2000)
Place of origin	Hamilton, Ontario, Canada
Language	English & Canadian French. Other translated versions also reported (French, Spanish-American, German, Dutch & Japanese) (www.fhs.mcmaster.ca/hug)
Purpose of measure	The HUI is a value & utility measure to assess preference scores for health states defined by a multiattribute health status classification. It determines a general public-based utility score for a specified health outcome or for the health status of an individual. In clinical populations, the scores can be used to provide a single summary measure of HRQoL or as quality weights for calculating QALYs in cost-utility analysis. In general populations, the measure can be used as quality weights for determining population health expectancy. The measure focuses on functional capacity rather than performance.
Item generation	Theoretical – multiattribute utility theory, evidence from the literature led to the development of the attributes
Respondent type	HUI II: Proxy – parent or clinician; child > 8 years of age HUI III: Proxy – parent for children; self-report for adults
Age range	HUI II: 2-18 years HUI III: birth-old age
Number of items	HUI II: 7 HUI III: 8 The HUI II & III may be combined to form a 15-item questionnaire
Number of dimensions (type)	HUI: 4 (physical function, role function, social-emotion function & health problems) HUI II: 7 (sensation, mobility, emotion, cognition, self-care, pain & fertility) HUI III: 8 (vision, hearing, speech, ambulation, dexterity, emotion, cognition & pain/discomfort)
Rating scale	HUI: 4-6 levels of functioning per attribute (960 health states) HUI II: 3-5 levels of functioning per attribute (24,000 health states) HUI III: 5-6 levels of functioning per attribute (972,000 health states)
Scale of studies	HUI I: 1) 87 children (Torrance et al, 1995); 2) 84 child/parent pairs (Cadman et al, 1986) HUI II: 1) 293 children (Torrance et al, 1996); 2) 28 & 13 children (Feeny et al, 1992); 3) 156 & 145 children (Saigal et al, 1994a & 1994b); 4) 10 children (Barr et al; 1994); 5) 48 children (Billson & Walker, 1994); 6) 61 children (Trudel et al (1998); 7) 468 children (Gemke & Bonsel, 1996; Gemke et al, 1995) 8) 18 children (Barr et al, 1997); 9) 27 children (Glaser et al, 1997a); 10) 47 children (Glaser et al, 1997b); 11) 141 & 123 children (Saigal et al, 1998a); 12) 30 children (Glaser et al, 1999); 13) 244 children (Speechley et al, 1999); 14) 42 children (Le Gales et al, 1999); 15) 80 children (Szechet et al, 1999); 16) 44 children (Barr et al, 1999); 17) 22 children (Mulhern, 1999); 18) 142 children (Felder-Puig et al, 2000)

Table 2.12 Summary review of the HUI I/II/III measures (Measure 9)

Name of measure	HUI (continued)
Sample composition	HUI I: 1) Parents of 87 school-aged children; 2) parents and children (aged grade 7-8) (Canadian study) HUI II: 1) 293 parents of school children in Ontario (Canada); 2) 20 children on & 8 children off therapy for acute lymphoblastic leukaemia, Wilm's tumour or neuroblastoma (Canadian study); 3) 156 survivors of NIC evaluated at age 8 years & 145 school children aged 8 years; 4) 10 parents of children with brain tumours (Canadian study); 5) 48 child/parent/doctor pairs (children aged 2-17 years; self-completed in children >8 years or parent-completed in children < 8 yrs) (UK study); 6) 61 mothers of paediatric oncology patients (20 on & 41 off treatment) (French study); 7) 468 Dutch children on a PICU; 8) 18 children receiving maintenance treatment for ALL (self-completed by children, also assessed by parent and health professional) (Canadian study); 9) 27 children aged 6-17 years with central nervous system tumours, 21 parents & 27 teachers (UK study); 10) 47 child/parent pairs (children aged 5-16 years) (UK study); 11) 141 ELBW survivors & 123 matched controls aged 12-16 yrs (self-completed by teenagers, also completed by parents) (Canadian study); 12) 28 children aged 6-16 years with cancer, 30 parents, & clinician and physiotherapist (UK study); 13) parents of 244 children aged ≥ 5 years with cancer (Canadian study); 14) 42 children with cancer aged 5-19 years (self-completed in children > 10 yrs, parent-complete < 10 yrs) (French study); 15) 80 children with cancer aged > 5 years (Argentina study); 16) 44 children with brain tumours & cognitive impairment (self-complete where possible, parent, nurse and doctor also reported health status) (Canadian study); 17) 22 children treated for medulloblastoma (US study); 18) 142 childhood cancer survivors aged 6-30 years (self-completed in children > 13 yrs, parent-complete < 13 yrs, all children assessed by health care professional) (Austrian study)
Reliability	HUI II: test-retest reliability is reported in childhood cancer and PIC studies (Barr et al, 1994; Gemke & Bonsel, 1996; Trudel et al, 1998; Glaser et al, 1999). Gemke & Bosel (1996) report high inter-rater reliability for domain scores with correlations generally above 0.8 among three different observers – parent, investigator and clinician. Barr et al (1994) reported correlations of 0.57-0.90 for global utility scores based on health status classification of paediatric brain tumour patients by clinicians, a nurse and parents. Trudel et al (1998) reported a weighted Kappa of 0.40-0.75 in their assessment of test-retest reliability. Reliability of the scoring mechanism in childhood cancer studies was proven with scores almost identical to the reference population (Torrance et al, 1995 & 1996), & good inter-rater reliability in childhood cancer studies (Glaser et al, 1997a & 1997b). Saigal et al (1998a) observed differences in emotion scores between parents and teenagers.
Validity	HUI II – Discriminant validity is reported in child studies (Feeny et al, 1992; Saigal et al, 1994a & 1994b; Trudel et al, 1998). Concurrent validity of HUI II & III tested with the CHQ (correlations of 0.45-0.64) (Speechley et al, 1999)
Responsiveness	Reported in one small study; HUI II global utility demonstrated moderate responsiveness with a ICC of 0.43 (Barr et al, 1997)
Practicality	HUI II: few minutes, self-administered questionnaire
Quality score	8 (reliability and utility = 2; validity, practicality, feasibility and user-centredness =1; responsiveness =0)

* Also used Child Health Questionnaire (CHQ)

Table 2.12 (cont.) Summary review of the HUI I/II/III measures (Measure 9)

The content validity of the HUI measure is however questionable. Glaser et al (1999) modified the wording of two items of the original HUI in their UK study but concluded that extreme caution is required if minor changes in the wording of questionnaires are

made as the weighting and interpretation of items may be altered significantly; such modifications are inadvisable (Glaser et al, 1999). Trudel et al (1998) also provided additional explanations to clarify item wording for parents of young children. Trudel et al (1998) reported the content validity of the HUI II to be more or less adequate as a descriptive health profile in children with cancer, following a review of the literature and gathering of experts' opinion. It covers important aspects such as emotion, but it is insufficient to evaluate essential domains such as neuropsychological and psychosocial functioning thoroughly (Trudel et al, 1998). Billson and Walker (1994) also modified the original HUI measure to ease comprehension for parents and children.

Criterion validity of the multiattribute utility function is reported as determining the ability of the model to predict directly measured scores not used to create the system (the criteria) (Torrance et al, 1995 & 1996). Convergent validity for the HUI II attributes of emotion, pain and self-care with other established measures are reported with low to moderate convergent validity (Trudel et al, 1998). The convergent validity of the HUI II utility score has shown a significant but low correlation with a five-point Likert scale assessing general health status (Trudel et al, 1998). Construct (discriminant) validity is reported with evidence that the measure can be used to distinguish between groups known to have clinically important differences in health status (Feeny et al, 1995). Studies report differences in health status and HRQoL between survivors of extremely low-birthweight at the age of eight years, and a group of reference children matched for age, gender and socio-economic status and control children aged eight years (Saigal et al, 1994a & 1994b). The HUI II also differentiates levels of morbidity between children with cancer who are on or off treatment (Feeny et al, 1992; Trudel et al, 1998). There is limited evidence of the responsiveness of the HUI measure (Barr et al, 1997).

Gemke & Bonsel (1996) reported good test-retest reliability results despite confounding by methods of administration, written versus verbal self-report. Test-retest reliability of the HUI Mark III system is also reported in an adult study only (Torrance et al, 1995). Practicality of the HUI II/III measures is reported favourably; the measures are relatively short and self-completion is estimated at several minutes. Glaser et al (1999) report the HUI as acceptable to respondents because of high completion rates in their study (93% children and 100% parents), but suggest that instructions should be clarified with respondents to reduce errors in completion. Children over seven years of age are reported

to complete the measure consistently (Juniper et al, 1997); Saigal et al (1998a) state children as young as eight years of age can provide reliable assessments of HRQoL. However, Saigal et al (1998b) were unable to apply the 1992 HUI classification system developed for school-age children to pre-school children because of differences in cognitive, self-care, and behavioural characteristics. They report a multiattribute pre-school health status classification for parent-completion describing functional health status of three-year olds; comprising twelve dimensions - vision, hearing, speech, mobility, dexterity, self-care, emotion, learning and remembering, thinking and problem-solving, pain, general health, and behaviour, with between 3-5 levels of functional limitation for each dimension (Saigal et al, 1998b).

The HUI II measure has been extensively used with some success in childhood cancer studies (Feeny et al, 1992 & 1993; Barr et al, 1994 & 1999; Billson & Walker, 1994; Glaser et al, 1997a & 1997b; Trudel et al, 1998; Glaser et al, 1999, Le Gales et al, 1999; Speechley et al, 1999; Szecket et al, 1999; Felder-Puig et al, 2000), and children admitted to intensive care (Gemke & Bonsel, 1996). The use of the HUI measure in children under five years of age is not well reported (Billson & Walker, 1994; Gemke et al, 1995; Gemke & Bonsel, 1996). The HUI measure appears reliable and valid but its content validity is weak; it encompasses the age-range of children in this research programme and was considered the best available 'gold standard' measure for the assessment of criterion validity.

Measure 10: Autoquestionnaire Enfant Imagé (AUQEI) Questionnaire

Three papers were identified on the AUQEI measure describing five studies (Table 2.13).

There is very little data described in the published papers to support the validity and reliability of the AUQEI measure; responsiveness was not tested. It is unclear how the AUQEI items were generated. An English version of the measure is available, but the measure was developed in a French population; therefore different dimensions may be identified in a UK population.

Name of measure	AUQEI questionnaire
References	Manificat et al (1995); Manificat & Dazord (1998); Manificat et al (1999)
Place of origin	Lyon, France
Language	French, but English & Spanish versions also available
Purpose of measure	Unclear, other than to measure a child's QoL using a profile approach
Item generation	Not reported, other than developed in a French population
Respondent type	Self-complete by the child, unless the child cannot read, in which case a "helper" is needed. Proxy - parent & clinician
Age range	4-12 years (Manificat et al, 1999)
Number of items	Version 1 – 9 items Version 2 – 27 items
Number of dimensions (type)	8 (family life & relations, external activities (schooling, sport), leisure activities, self-image, separation situations, major functions, social relations, & treatment situations)
Rating scale	Version 1 - child indicates their level of satisfaction in each dimension & is asked to name typical situations associated with emotional states; global indices Version 2 - profiles for the structured formatted satisfaction scales. Child's level of satisfaction with each dimension is measured along a 4-level response continuum represented by faces indicating level of satisfaction (Manificat & Dazord, 1998). The child also relates a situation from his/her own experience to each of the faces representing a satisfaction level, & is asked to specify how often the pleasant or unpleasant situation happens (Manificat et al. 1995).
Scale of studies	Version 1 - tested in 2 successive samples of 54 & 71 children in a hospital setting Version 2 – tested in 3 populations of children: 1) 102 children; 2) 169 children; 3) 40 children
Sample composition	Version 1 - 59% of the 54 children suffered from a serious disorder (mean age was 9.2 yrs, minimum age was 5 years); of the 71 children, half were in good health, half were under medical treatment in hospital – 14 Human Immunodeficiency Virus (HIV) positive, 22 with kidney transplants Version 2: 1) 60% healthy, 26% renal disease & 14% HIV positive; 2) 102 children were incorporated from the 1st sample plus 53 children with haematological diseases & 14 children with affective disorders; 3) 40 children with psychosocial problems in residential placement
Reliability	Internal consistency of Version 1 reported with $\alpha = 0.51$ and $\alpha = 0.71$ for Version 2. Poor inter-rater reliability reported between parents and doctors (0.3). Test-retest reliability not reported.
Validity	Criterion validity (Version II) assessed by relating scores to that of a third party responsible for the child's treatment - Spearman rho coefficients of 0.30 ($p<0.01$) reported (Manificat & Dazord, 1998). Discriminant validity was tested with comparative studies between healthy & sick children; certain items discriminated better than others (sensitivity). Construct validity tested (Version II) via principal components analysis (PCA) producing 4 factors: autonomy; leisure activities (holidays, birthdays); "functions" (sleep, meals, school); and parents (Manificat et al, 1995).
Responsiveness	Not tested
Practicality	Children understood Version I items; completion time not reported
Quality score	3 (validity, user-centredness and utility =1; reliability, responsiveness, practicality and feasibility =0)

Table 2.13 Summary review of the AUQEI questionnaire (Measure 10)

Measure 11: PedsQL™ 4.0 (Pediatric Quality of Life)

Six papers describing three studies were identified on the PedsQL measure; five of these papers were reviewed as two described the same study but in different publications* (Table 2.14).

Name of measure	PedsQL
References	http: www.pedsql.org (Varni, 1998); Varni et al (1998a & 1998b); Varni et al (1999a); Varni et al (1999c); Varni et al (1999d)*; Seid et al (1999a)*; Seid et al (1999b)†
Place of origin	USA
Language	English, also translated into Spanish & Arabic, with German & Dutch in progress
Purpose of measure	Generic Scales – designed to enable comparisons across patient & healthy populations (Varni et al, 1999d) Disease-Specific Modules - designed to provide greater measurement sensitivity for circumscribed clinical populations (e.g. asthma, arthritis, cancer, cardiac disease & diabetes) The measures can be used to assess risk; to track health status, & to monitor treatment outcomes in patients most likely to be seen in paediatric tertiary care facilities.
Item generation	Developed from earlier measures PCQL & PCQL-32 for use in childhood cancer populations. Items generated from literature reviews, open-ended interviews with patients & their families, & discussions with health care professionals (nurses, specialists, & psychosocial staff).
Respondent type	Proxy – parent (2-18 years); child complete (5-18 yrs)
Age range	2- 18 years Age-appropriate forms for children aged: 2- 4 yrs; 5-7 yrs; 8-12 yrs; & 13-18 yrs
Number of items	Generic Core Scales - 23 items
Number of dimensions (type)	Generic Core Scales – 4: (physical functioning - 8 items; emotional functioning - 5 items; social functioning - 5 items; & school functioning - 5 items)
Rating scale	3-point Likert scale (5-7 year-olds); 5-point Likert scale (8-18 year-olds) Total scale score (0-100), physical health summary score & psychosocial health summary scores are calculated
Scale of studies	1) PedsQL 4.0 - Generic Core Scales: 1511 children & adolescents – 820 self-reports for children aged 5-18 years, & proxy-report for 1476 parents of children aged 2-18 years (785 cases of child and parent-report) (Varni et al, 1999b) 2) PedsQL – 291 children with cancer & their parents (8-18 years old) (Varni et al, 1999d) 3) PedsQL-32 – 325 children with cancer & their parents (8-18 years) (Varni et al 1998a & 1998b; Varni et al, 1999a)
Sample composition	1) 6.5% healthy children at routine preventive paediatrician visits; acute & chronically ill children seen in orthopaedic (8.4%) & cardiology (10%) specialty clinics; 75% (638 healthy & 481 chronically ill children) seen as out-patient, in-patient or emergency patient at Children's Hospital & Health Centre. 2) Newly diagnosed children on-treatment, relapsed on treatment, in recent remission off treatment & long-term off treatment (mainly children with acute lymphoblastic leukaemia) 3) As above; 13 different cancer diagnoses
Reliability	Internal consistency reported for PCQL-32, PedsQL & PedsQL 4.0. The reliability of the PedsQL 4.0 is reported as $\alpha > 0.7$ & approaching 0.9 for the full 23-item scale for self and proxy-report (Varni et al, 1999b). The reliability for the total scale score is reported as 0.88 child self-report, and 0.90 for parent proxy-report (http: www.pedsql.org/about_pedsql.html, 2nd August 2000).

Table 2.14 Summary review of the PedsQL (Measure 11)

Name of measure	PedsQL (continued)
Validity	PCQL-32: convergent validity tested against other measures of disease burden; discriminant validity reported; clinical validity tested via known group comparisons. PedsQL: construct and clinical validity reported. PedsQL 4.0: discriminant validity of the generic core scale is reported via known groups comparison & correlation with other measures of disease burden. Results report that the self-report & proxy-report measures distinguished between children with & without a chronic health condition, between those who did or did not have an overnight hospital visit in the last twelve months (Varni et al, 1998b). Significant correlations were found with other measures of disease burden.
Responsiveness	Responsiveness of PedsQL 4.0 reported but no evidence
Practicality	PedsQL 4.0 Generic Core Scale – age-appropriate form takes less than 4 minutes to complete; PCQL-32 administered by research assistant
Quality score	5 (reliability, validity, practicality, feasibility and utility = 1, responsiveness and user-centredness = 0)

* Describes the same study, only Varni et al (1999d) reviewed.
† Identified by 2nd reviewer – abstract not found.

Table 2.14 (cont.) Summary review of the PedsQL (Measure 11)

The PedsQL version 4.0 builds upon and expands a programmatic measure development effort by Varni and colleagues, from the USA, during the past 15 years in paediatric populations. Version 1.0 was originally derived from a paediatric cancer database, which was designed as a generic QoL inventory to be utilised non-categorically, i.e. across multiple paediatric populations in children aged 8-18 years (Varni et al, 1998a & b). The PedsQL 2.0 and 3.0 were further advancements in the measurement model, including additional constructs and items, a more sensitive scaling range, and a broader age range for patient self-report and proxy-report (Varni et al, 1999c).

The PedsQL version 4.0 is a multidimensional measure, which uses a modular approach comprising generic and condition-specific modules (Varni et al, 1999b). The authors report that the use of version 4.0 in clinical trials is in the planning stages, but the measure has been field tested in paediatrician's offices, speciality clinics, and community settings. The Generic Core Scales are currently in use in several school districts and State Departments of Health – to monitor the health of large populations of healthy and ill children (Varni et al, 1999a).

The PedsQL version 4.0 Generic Core Scales are reported as being tested in physically healthy paediatric populations and in acutely and chronically ill children. Internal consistency of the measure is reported as excellent but evidence to substantiate this statement is not provided. The test-retest reliability of the measure is not reported. Discriminant validity is reported, but data to substantiate these findings are not described

but 'in press'. The authors describe the measure as responsive to clinical change as demonstrated in a recent field trial (results in press), but no evidence is provided (Varni et al, 1999b). Practicality of the measure is supported with the Generic Core Scales printed on one side of paper, with instructions on the reverse, and completion time taking a few minutes.

Much of the information on the PedsQL 4.0 measure is still in press and therefore unpublished; one paper alone describes the development of the PedsQL 4.0 measure in healthy, and acutely and chronically ill children. The authors report the psychometric properties of the PedsQL 4.0 measure favourably, but supporting information is not publicly available, except via an Internet web page. There is no published information available on the psychometric properties of the disease-specific modules.

The PedsQL 4.0 measure was developed from earlier measures that were specifically developed in children with cancer, the PCQL-32 (Varni et al 1998a & 1998b; Varni et al, 1999a), and the PedsQL measures (Varni et al, 1999d). No published evidence is available on the psychometric properties of versions 2.0 and 3.0 of the PedsQL measure. The age-appropriate forms of the PedsQL 4.0 seem a good method of addressing the cognitive development issues when developing a measure for children, but the measure has not been validated in children less than two years of age. This forms a large proportion of children admitted to PIC. For this reason and because of the limited published information on its psychometric properties, the measure was not considered for use in this research programme.

Measure 12: QUALIN - Infant Quality of Life Questionnaire

One short paper (same paper as AUQEI measure) was identified on the QUALIN measure with little evidence provided by the authors to substantiate their findings (Table 2.15).

The authors state that their questionnaire appears satisfactory in terms of validity and specificity, and internal consistency; similar results were obtained for each country (Manificat et al, 1999). Low correlations were obtained in the assessment of criterion validity; it was unclear if the measure was administered at the same time to both types of respondent in this assessment. Response rates were good, but no information was provided on questionnaire completion times.

Name of measure	QUALIN
References	Manificat et al (1999)* (*also described in Measure 10)
Place of origin	Lyon, France
Language	French, but also translated into Spanish & Italian
Purpose of measure	To assess QoL in infants < 3 years of age
Item generation	Survey of 800 parents/caregivers in France regarding their answer to the question, "what allows you to say that your child has a good or bad quality of life? Please list 5 components which contribute to your infant quality of life." A content analysis of these responses led to the development of a structured formatted scale.
Respondent type	Proxy - parent & caregiver (clinician)
Age range	Children under 3 years of age Form 1: < 1 year Form 2: 1-3 years
Number of items	33
Number of dimensions (type)	Unclear (examples include somatic, sociability, environment, separation, & psychopathology)
Rating scale	5-point response scale from -2 (definitely false) to +2 (definitely true). A global score can be computed by adding the individual scores obtained for each of the items or from the different factors found in the principal components analysis.
Scale of studies	1) Preliminary parental survey (n=800) to develop items. 2) 1433 children (90% response rate). Paediatricians also completed 70 questionnaires (80% response rate)
Sample composition	2) Children from 6 countries (France, Belgium, Luxembourg, Switzerland, Spain and Italy); 50% were children were <1 year, & 50 % were aged 1-3 years (equal number of boys & girls); a few children had chronic illnesses, 22-36% had an acute illness, 85% of children were out-patients in private surgery.
Reliability	Internal consistency reported, $\alpha = 0.76$-0.80
Validity	Concurrent validity revealed significant correlations (0.40) for 29 of 33 QoL items between parents and paediatricians, with less significant correlations for children <1 year of age. Parents scored more optimistically than paediatricians for both forms, except for items relating to psychological or somatic items, where there was no difference Construct validity was assessed via PCA: the emerging factors were in agreement with the hypotheses linked to the questionnaire construction, but differed between form versions
Responsiveness	Not reported
Practicality	Reported as response rate only
Quality score	4 (reliability, validity, practicality and utility = 1; responsiveness, feasibility and user-centredness = 0)

Table 2.15 Summary review of the QUALIN measure (Measure 12)

The QUALIN measure encompasses children aged 0-3 years, most of the specified age-range of children in this research programme, but the psychometric properties of content validity, responsiveness and test-retest reliability were not described. It is unclear how the questionnaire was devised, or the exact purpose of the measure, without referring to additional papers published in French. This measure was devised in a French population; there is currently no English translation available. Translating this measure into English would be resource intensive and is not warranted in this research programme.

Measure 13: Warwick Child Health & Morbidity Profile (WCHMP)

One paper identified three studies on the WCHMP measure, which was developed over three phases (Table 2.16).

Name of measure	WCHMP
References	Spencer & Coe (1996)
Place of origin	Coventry, UK
Language	English
Purpose of measure	To measure parent-reported health & morbidity in infancy & childhood. It is suitable for research & service planning purposes & is capable of measuring both cross-sectional & longitudinal health & morbidity experience in a child population. It is not a health index.
Item generation	Not reported
Respondent type	Proxy-parent
Age range	0-5 years
Number of items	16
Number of dimensions (type)	10 (general health status; acute minor illness status; behavioural status; accident status; acute significant illness status; hospital admission status; immunisation status; chronic illness status; functional health status; & HRQoL)
Rating scale	Each dimension consists of a single global question with four categories of response; dimensions are not weighted or scored. Details of acute minor illness, behavioural problems, accidents, hospital admissions, acute significant illness and chronic illness were obtained using second tier questions.
Scale of studies	228 children in 3 phases: 1) 128 parents 2) 60 parent 3) 40 parents
Sample composition	'Normal' pre-school children & those with developmental problems & acute & chronic illnesses 1) 47 in child health clinics, 30 in child development units & 51 in out-patient departments in Coventry (open-questions asked of 20 parents) 2) 20 in each setting as detailed above 3) Measure administered twice to 40 parents during routine child health centre visits, once buy investigator, once by health visitor
Reliability	Inter-rater reliability (investigator and health visitor) was good with weighted Kappa statistics ranging from good to very good (0.76-1.00). Test-retest reliability (2 weeks - 3 months) reported in 88 out of 128 interviews using weighted kappa statistic (0.50-0.86); varied from moderate to good to very good depending on the dimension of health assessed - life quality status was reported as moderate.
Validity	Criterion validity was assessed via medical judgements from two paediatricians, based on parental responses to second tier questions. Results showed a weighted Kappa statistic ranging from good to very good (0.770-0.949); and parental recall against medical records which showed high correlations with the health records. Construct validity was tested by exploring the data to test compliance with four medical constructs of child health; results revealed that the relationships between variables predicted by all constructs occurred more frequently than expected by chance at the 95% level.
Responsiveness	Not reported
Practicality	10 minutes to complete
Quality score	6 (reliability, validity, practicality, feasibility, user-centredness and utility =1; responsiveness = 0)

Table 2.16 Summary review of the WCHMP measure (Measure 13)

The WCHMP is not a multidimensional measure of HRQoL as it focuses more on the measurement of morbidity. Many of the items focus on health care resource use with only two items measuring functional status and HRQoL. The dimensions are assumed to be clinician-generated with minimal input from parents other than via open-ended questions within the measure. These dimensions may not be considered to be the most important to parents.

The psychometric properties of the measure are reported favourably, but responsiveness was not tested. The measure is suitable for children aged 0-5 years, but was not considered for use in the research programme because of its purpose and the limited items on HRQoL.

2.5 Quality scores

Scores for all seven criteria (reliability, validity, responsiveness, practicality, feasibility, user-centredness, and clinical utility) ranged from 2-8 (maximum score = 14); scores for four criteria (reliability, validity, responsiveness and practicality) ranged from 0-4 (maximum score = 8) (Table 2.17).

Name of measure	Quality score (7 criteria)	Quality score (4 criteria)
CAQ	7	4
RAND HIE I & II	5	2
CHSI for Ontario Children	5	2
NHIS	2	0
FS II (R)	6	4
QWB Scale	5	4
Nordic QoL Questionnaire	5	2
ITQoL Questionnaire	0	0
HUI	8	4
AUQEI	3	1
PedsQL	5	3
QUALIN	4	3
WCHMP	6	3

Table 2.17 Summary of the quality scores of included child HRQoL measures

Streiner and Norman (1995) state that outcome measures should meet the criteria of reliability, validity, responsiveness and practicality; none of the measures reviewed met all four criteria. Six measures partially fulfilled the criteria for reliability and validity (scoring 1 for each); one measure partially fulfilled the criteria for reliability (scoring 1) but met the

criteria for validity (scoring 2); and two measures met the criteria for reliability (scoring 2) and partially met the criteria for validity (scoring 1). One measure scored zero, the ITQoL measure, of which there was no published information available.

2.6 Discussion

The review identified 51 papers describing thirteen measures for use in children aged 0-5 years: twelve generic and one respiratory-specific. Guidelines described by Feinstein (1987) and Greenhalgh et al (1998) were utilised to inform judgements about the validity and reliability of the measures identified. The criterion of reliability, validity, responsiveness, practicality, feasibility, user-centredness and clinical utility were assessed for each measure. The quality scores for the twelve generic and one respiratory-specific measures were generally poor, ranging from 2-8 (maximum score possible = 14). When considering the four essential criteria of measures described by Streiner and Norman (1995), no measures met all criteria, and only six measures partially met the reliability and validity criteria. The low quality scores identified for the included measures could reflect the difficulties in developing child HRQoL measures, the limited published evidence about their psychometric properties, and the embryonic development of most of these measures. Many of the papers described validation studies (field trials) with very few papers describing the use of the HRQoL measures in routine clinical practice, with the exception of the HUI measure.

This review was carried out rigorously; publication bias was minimised via hand searching and reviewing grey literature. Reviewer bias was minimal with the search procedure and quality scoring system revealing reliable results. Utilising the second reviewer to assess titles obtained from initial electronic searches to identify papers to include in the review may have enhanced the reliability of the review, but was beyond the financial and time resources available.

The review identified 47 studies and differs from findings in other reviews because it identified more measures and used a quality score to rate the psychometric properties of the identified measures. Eighteen generic and three respiratory-specific HRQoL measures were excluded, as they were only appropriate for children over the age of five years (Appendix II). The review described six measures for children aged 0-5 years and five measures for children over five years of age that were not described by Eiser and Morse

(2001a). Eiser and Morse (2001a) identified the Perceived Illness Experience measure as one of nineteen generic measures in their review. This measure was initially developed to assess perceived illness experience in young people with cancer (aged 8-18 years), but may be used with other groups of children or young people with chronic illness (Eiser et al, 1995; Eiser et al, 1999). This measure was excluded from this systematic review, as it is not a measure of HRQoL per se.

2.6.1 Implications of the review for the development of the HRQoL measure

This review failed to identify a suitable HRQoL measure to evaluate HRQoL outcomes in children under the age of five years, with a respiratory illness, in a PIC setting. However, the review informed the selection of the best available 'gold standard' measure to include within the research programme to assess criterion validity in a new HRQoL measure. Selection was based upon the strength of the psychometric properties of the measure and its age applicability.

The review identified twelve generic HRQoL measures applicable to the age-range of interest in this research programme; however, the minimum age of three of these measures was four years, and two years for two other measures. Most children who receive PIC are under the age of two years; the choice of HRQoL measure needed to encompass this age group. Six measures reported a minimum age of zero; one of these measures, the HUI, reported an eligible age ranging from birth or two years of age to old age. Of six HRQoL measures identified that covered the entire age-range of interest (0-5 years), two measures were strongly considered as the best available 'gold standard' measure: the HUI and ITQoL measure. Four other measures (RAND HIE, NHIS, FS II (R), and WCHMP) were discounted owing to the lack of evidence on their psychometric properties and the purpose for which one measure (WCHMP) was designed. The CAQ measure (measure 13) was also discounted because of its high minimum age limit (4 years), and because its psychometric properties would need further testing in children with respiratory illnesses other than asthma.

The strengths of the HUI and ITQoL measures were that they encompassed the age-range of the PIC population to be studied in the research programme. The ITQoL measure was developed specifically for infants and toddlers aged two months to four years. The HUI measure has been used extensively in adult and paediatric studies, with the paediatric

studies mainly comprising children with differing types of cancer (Feeny et al, 1993; Barr et al, 1994; Trudel et al, 1998); one study is reported in a PIC population (Gemke et al, 1995; Gemke & Bonsel, 1996). The HUI measure is relatively short and is reported to take only a few minutes to complete. Published information is also available on its psychometric properties, which are reported favourably.

The weaknesses of the ITQoL and HUI measures were that they were developed in American and Canadian child populations respectively, which are culturally different from a UK child population. The ITQoL measure is also long, comprising 104 items, which could be time-consuming for parents to complete alongside a newly developed HRQoL measure. Information about the use of the ITQoL measure is sparse, with limited published information available. Published information about the HRQoL measure was one of the inclusion criteria for use of a measure within the research programme. The content validity of the HUI measure is weak as it was developed in children over the age of five years. Modifications in the wording of HUI items have been reported, to clarify the items for use in young children. Despite these limitations, the HUI measure was selected as the best available 'gold standard' measure because of the stronger published evidence on its psychometric properties.

2.7 Summary

This chapter described the methodology and results of a methodological systematic review of child HRQoL measures including generic and respiratory-specific (disease-specific) measures, utilising published guidelines advocated by the NHS CRD (1996). The review was extensive and included a quality assessment of the included measures, but was limited by time and financial constraints. No 'gold standard' measure exists, but the review identified the best available 'gold standard' measure to test the criterion validity of the HRQoL measure to be developed in the research programme, the HUI Mark II and III versions. The findings of the review support those described by Eiser and Morse (2001a) but identified more specific measures. None of the measures included in this review evaluated all theoretical dimensions of health described in the literature.

CHAPTER 3 – Qualitative study to generate items to measure the parental perspective of HRQoL

3.0 Introduction

This chapter describes a study investigating parental perceptions of child health and HRQoL. A qualitative method of inquiry was employed using semi-structured interviews with parents of children discharged from PICU. The interview data was analysed using thematic content analysis. Leventhal's self-regulatory theory of illness representations formed a theoretical framework for the data analysis phase (Leventhal, Meyer & Nerenz, 1980; Leventhal, Nerenz & Steele, 1984; Leventhal & Nerenz, 1985). Themes identified from the analysis informed the development of parent-generated items for inclusion in the HRQoL measure.

3.1 Background

The purpose of HRQoL measures is to assess the views of the patients themselves to capture important aspects of health status and HRQoL from the patient perspective (Jenkinson & McGee, 1998) (Chapter 1). Streiner and Norman (1995) recommend in-depth interviews as a method of item generation with a small number of participants, such as patients and clinicians with unique knowledge. Patients' views are fundamental in helping to decide which treatments are best, given their own values and risk preferences.

Obtaining the patient perspective on HRQoL is problematic when the patients are children (Chapter 1). Parents are next best placed to assess HRQoL and are commonly used as major informants in child HRQoL assessments (Levi & Drotar, 1998). Parental reports therefore represent a proxy response about their children's function. Involving parents in the generation of items aims to identify their perceptions of the relevant aspects of health and HRQoL, thus strengthening the face and content validity of the HRQoL measure. Both face and content validity are a minimal prerequisite for acceptance of an HRQoL measure (Streiner & Norman, 1995).

There is little prior evidence as to what parents think about HRQoL or their beliefs about illness in PIC. However, in other areas, illness perceptions seem clustered around identity, cause, timeline, consequences and cure or control. These areas are described in a self-regulatory model of illness cognitions and representations (Leventhal et al, 1980;

Leventhal et al, 1984; Leventhal & Nerenz, 1985). Illness cognitions are defined as a patient's implicit commonsense beliefs about their illness (Leventhal et al, 1980; Leventhal et al, 1984; Leventhal & Nerenz, 1985); patient's illness representations are based around distinct components, which in turn determine coping (Weinman et al, 1996). The components are:

- *Identity* – (diagnosis / symptoms)
- *Cause* - the perceived cause of the illness (biological / psychological)
- *Timeline* – how long the illness will last
- *Consequences* – effects of the illness on a person's life
- *Curability and controllability* – illness can be cured and whether outcome is controllable by the person or powerful others.

3.1.1 Aim

This study aims to identify parental beliefs about health, QoL and PIC illness.

3.1.2 Research questions

The following research questions were formulated:

- What are parents' perceptions of their child's health and wellbeing following treatment on PICU for a respiratory illness?
- How do parents perceive the concept of child health and the characteristics of a healthy and unhealthy child?
- How do parents perceive the concept of QoL and the characteristics of a child with a good and poor QoL?
- What factors influence parental perceptions of child health and QoL?

3.2 Methods

3.2.1 Design

This study employed cross-sectional, semi-structured interviews as a method for exploring the new area of parental perceptions of health and HRQoL. Qualitative research methods can reach attitudes and interactions that quantitative methods cannot by developing concepts which help the understanding of social phenomena in natural settings, and giving due emphasis to the meanings, experiences, and views of all the participants (Cobb & Hagemaster, 1987; Creswell, 1994; Schwandt, 1994; Mays & Pope 1996a). An exploratory

qualitative research approach also allows the exploration of concepts to find out what is happening, to seek new insights, to ask questions, to assess phenomena in a new light, or challenge the status quo, and obtain insightful and rich data on complex issues (Robson, 1995; Bowling, 1997b). Qualitative methods are also essential in the initial stages of questionnaire design and scale construction, and can supplement quantitative work as part of a validation process, or through exploring areas not amenable to quantitative research (Mays & Pope, 1996a; Bowling, 1997b; Russell, 1998). When used jointly, qualitative and quantitative methods can combine to form a powerful research tool (Polgar & Thomas, 1998). The exploratory nature of the research questions in this study meant that a qualitative research design was justified. Comparisons or relationships were not the main focus of the study; this approach lends itself to a quantitative research design.

Semi-structured interviews, or respondent interviews, aimed to provide rich, in-depth data, on parental perceptions of health and HRQoL, despite being time-consuming to conduct. This data collection method has clear advantages for those parents who may have difficulty reading or writing (Fowler, 1996). Face-to-face interviews with parents allowed the interviewer to remain in control of the interview with the possibility of modifying the line of enquiry and enabling non-verbal cues to give messages to help in the understanding of the verbal response (Robson, 1995, Mason, 1996). Conducting interviews requires skill and experience in technique (Robson, 1995). It is reported that detailed explanation of some questions and the flexibility of semi-structured interviews may introduce bias as the interview may become more conversational and less structured (Harris & Inayat, 1997); bias is difficult to rule out if probes are used (Oppenheim, 1992). Interviewer bias was minimised in this study by reading out explanatory instructions to all parents.

3.2.2 Sample

The sample comprised main carers, referred to as parents, of children admitted to a PICU. The PICU at the research site is geographically split between two sites. The larger unit (10 beds) specialises in the care of critically ill children with neurological illnesses, and is a regional centre. The smaller unit (2 beds) specialises in the care of children with liver and renal disease, and childhood cancers. Children were sampled from the larger unit in equal groups from two points in time, three months (1999) and twelve months (1998) post PICU discharge.

3.2.2.1 Sample selection

The sample was selected from the sampling frame according to the following inclusion criteria:

- Children were sampled, not parents
- Primary PICU admission diagnosis was of a respiratory illness
- Child was < 5 years of age
- Child's PICU length of stay was ≥ 4 days
- Child was alive at time of sampling
- Child was resident within the Northern and Yorkshire region (North, West & East Yorkshire)
- Child's parents were English-speaking.

The sampling frame used was the PICU computer database, the accuracy and completeness of which was cross-referenced with the ward admissions book, a secondary sampling frame. A single-stage sampling procedure was used, as the names of children in the population were available to the researcher (Creswell, 1994). The sampling frame may have contained missing elements, that is children who failed to be entered on the database, or those children admitted to PICU for a short period of time before transfer elsewhere. The PICU ward admissions book was utilised to minimise the problem of missing elements (Moser & Kalton, 1996). Foreign (blank) elements included those children who died and those children aged over five years; these elements were ignored when selected (Moser & Kalton, 1996). Duplicate elements existed where the same child was admitted more than once in the timeframe specified for the study. In this situation, unique identification represented one of the admissions, and the others were treated as blanks (Moser & Kalton, 1996). A cluster of elements was not a problem in this sampling frame as the children were identified as individuals.

A PICU length of stay of four days or more was chosen as this represents the mean length of stay for children admitted to PICU. It was deemed inappropriate to interview bereaved parents in this study as this may have resulted in unnecessary parental distress and anxiety. Children aged 0-5 years were chosen, as this is the most common age group of children admitted to PICU; children with respiratory illnesses form the largest diagnostic group (Chapter 1).

3.2.2.2 Sample representativeness

Children were randomly selected from the sampling frame so that each child in the sample had an equal probability of being selected (Creswell, 1994). A random selection procedure is most rigorous, enabling the generalisation of the study findings to the entire population (Creswell, 1994). A random sample was chosen to obtain a representative sample of children with respiratory illnesses, and to provide raw data for comparative analysis (Morse, 1991; Mays & Pope, 1996a). Random number tables were used to avoid any bias owing to possible ordering of the sample (Moser & Kalton, 1996).

3.2.2.3 Sample size

For qualitative studies between 6-20 participants are thought to be adequate to generate items, especially if a representative sample is used (Harris & Inayat, 1997). Large qualitative studies do not often interview more than 50 or 60 people (Britten, 1996).

3.2.3 Researchers' role

In qualitative research, the role of the researcher as the primary data collection instrument necessitates the identification of personal values, assumptions and bias at the outset of the study (Creswell, 1994). The researcher (author) had experience of paediatric nursing, and intensive care nursing; thus, the researchers' understanding of the research context and role as a paediatric nurse aimed to enhance awareness, knowledge and sensitivity to the many issues encountered by parents whose children were treated in intensive care. The study was commenced with the perspective that parental perceptions of health and HRQoL are diverse and influenced by a number of factors, including parental experience of health, illness and children, and the researchers' experience of nursing. Distinguishing between the role of researcher and paediatric nurse may be difficult in situations where parents are distressed, and may introduce bias. Researcher bias may also be a problem, as a researcher may tend to selectively observe and record certain data at the expense of other data (Stern, 1991).

Researcher bias was minimised by referring distressed parents to the PICU family care nurses, and audiotaping the interviews, transcribing the interviews verbatim, inviting parents to review a copy of their interview transcript, and exposing the data analysis procedures to an expert in qualitative data analysis (Brink, 1991). The coding schedule

was checked with advice from research advisory group (RAG) members on neutral questions not associated with care.

3.2.4 Materials

A semi-structured interview schedule was developed using the research questions as a framework. This enabled a series of questions to be worked out in advance, with the freedom to modify the order based upon what seemed appropriate during the conversation, the wording of questions could also be changed or further explanations given, or particular questions left out which seemed inappropriate to a particular interview and additional ones included (Robson, 1995). Interviews thus allow flexibility.

The interview schedule was designed to start with questions that parents could answer easily, such as questions about the child's PICU admission, before proceeding to more difficult or sensitive questions, such as views on child health and HRQoL (Britten, 1996). The interview schedule included introductory comments, list of key questions to ask under topic headings, a set of associated prompts, and closing comments (Robson, 1995). Open-ended questions were used to probe, clear up any misunderstandings, test the limits of parental knowledge, and encourage co-operation and rapport. This approach also promoted unexpected or unanticipated answers, which may suggest new relationships or hypotheses; thus, permitting theory generation or interpretative analysis (Hutchinson & Skodol-Wilson, 1992; Yin, 1994). Open-ended questions, however, have the disadvantage of loss of control by the interviewer, and in particular in being more difficult to analyse than closed questions (Robson, 1995).

3.2.4.1 Ethics

Local Research Ethics Committee (LREC) approval was given in March 1999.

3.2.4.2 Piloting

The interview schedule was pre-tested to evaluate the questions, clarify wording and sequence the topics, and to reduce bias. Pre-testing the interview schedule resulted in additional explanations being incorporated as prompts within the schedule. These explanations were required to aid participant comprehension.

3.2.4.3 Coding frame – theoretical framework

The WHO definition of health and the dimensions of health identified in the systematic review informed the development of themes in the analysis of parental interview data (Chapters 1-2). Four interview transcripts were used to inform the coding frame. Some themes were informed by:

- Risk factors for respiratory disease
- Illness perceptions
- HRQoL

Other themes were informed by information not classified.

3.2.5 Procedure

General practitioners of selected children were contacted to confirm that the child was alive and well before parents were contacted by letter and invited to participate in the study. The family care nurse on the PICU was also contacted to confirm those parents whose children had died. The letter contained information on why, and for whom the interviews were being conducted; what was expected to emerge from the study; to whom the results were of interest to; and confidentiality and anonymity of the data (Moser & Kalton, 1996). Mothers were invited to participate in an interview at their home or in hospital, with or without their partner (Appendix III). Parents were invited to return an appointment slip detailing their preferred location, date and time of interview. The author contacted parents by telephone to arrange the interviews. Travelling expenses were reimbursed to parents who chose a hospital-based interview.

Prior to the interview, the purpose of the interview was described and an information sheet given to parents (Appendix III). Written informed consent was also obtained from parents, including permission to obtain information from other health care resources (Appendix III). Permission was also sought from parents to audiotape the interview to facilitate data analysis. Follow-up was also offered to parents by inviting them to obtain a summary of the research results by completing a section on the consent form (Appendix III). Notes were made 'in the field' following parental interviews to describe the context of the interview, observation of non-verbal communication during the interview, and observation of the child if present. A matrix was devised to collect brief demographic information on

the child and parent prior to the interview. This information was supplemented by details from the PICU ward admissions book.

3.2.6 Data analysis

The core of qualitative data analysis lies in the related processes of describing phenomena, classifying it, and seeing how concepts interconnect (Dey, 1993). Notes made 'in the field' were written-up to include self-reflection but were kept separately from the raw data and reviewed regularly (Robson, 1995). A full transcript of tape recordings was conducted and these were analysed simultaneously with data collection, data interpretation, and narrative report writing, using thematic content analysis (Creswell, 1994). Phrases were coded into categories (first-level coding), which included an examination of the context of the phrase in order to categorise it. Memos (analytical notes) were also written-up as a useful means of capturing ideas, views and intuitions at all stages of the data analysis process (Robson, 1995). Second-level or pattern coding was then performed to group the initial codes into a smaller number of themes or patterns (Robson, 1995).

Thematic content analysis allows the researcher to test theoretical issues to enhance understanding of the data and to develop an understanding of the meaning of communication (Cavanagh, 1997). Central to the methodology is the distillation, through analysis, of words into fewer content-related categories (Cavanagh, 1997). Content analysis is a procedure for categorising data for the purpose of classification, summarisation and tabulation (Cavanagh, 1997). Frequency counts were used to compare data from the content analysis and themes were ranked (Cavanagh, 1997).

A software tool for qualitative data analysis (NUD*IST 4.0 – Non-numerical Unstructured Data Indexing, Searching and Theory-building, Qualitative Solutions and Research (QSR) Pty Ltd, 1997) was used. The software package aids the management of documents, creation of ideas and management of categories and allows the asking of questions and building and testing of theories about the data – inductive analysis (QSR, 1997). The computer package provides an excellent medium for storing data and allowing easy access to it; it also provides procedures for coding data quickly and easily and is a powerful tool for searching data (Dey, 1993). Criticisms of computer-based analysis include the encouragement of data fragmentation so that instead of studying the data in-situ, the data are fragmented into bits and the overall sense of the data is lost (Dey, 1993). However,

content analysis can be extremely laborious and time-consuming and computerisation has led to substantial benefits (Robson, 1995). The computer software can also provide a category count, which is the central activity in content analysis (Robson, 1995).

The NUD*IST software is reported to have several advantages over manual systems including allowing the manipulation of the coding system to accommodate emerging themes, the ability to search for significant words or phrases, the ability to ask questions by including and excluding other nodes, to allow for comparison of questioning style and coding of a research team, and to carry out both inductive and deductive analysis (Pateman, 1998). The time taken to learn the package is a disadvantage, with a tendency to index anything and everything obsessively and unnecessarily (Pateman, 1998).

3.2.6.1 Validity and reliability checks of the data

The approach to qualitative data analysis needs to be rigorous and systematic. Four criteria need to be considered – credibility (analogous to 'internal validity'), transferability (analogous to 'external validity'), dependability (analogous to 'reliability'), and confirmability (analogous to 'objectivity') (Robson, 1995).

Credibility was addressed in the analysis by checking with those from whom the data was derived (Robson, 1995). Ten parents verified their interview transcripts as an accurate reflection of their interview; however, replies were not received from twelve parents. One parent suggested a slight amendment to their transcript concerning information about their child's illness. Peer debriefing was also utilised to establish credibility; data analysis and conclusions were exposed to the author's research supervisor to assist in the development of the design and analysis of the study (Robson, 1995).

Reproducibility is a form of inter-coder reliability and refers to the extent to which more than one coder independently classifies material in the same way as his or her colleagues (Cavanagh, 1997). It is important for a high degree of reproducibility to exist in content analysis, as this signifies a measure of shared, rather than individual, understanding of the data (Cavanagh, 1997). Precise coding instructions enhanced the reliability of the content analysis. Two parental interviews (10% of the sample) were independently coded by a second coder (research supervisor) using the thematic analytic framework to assign themes to the data. The degree of agreement or concordance between the two was calculated using

Cohen's Kappa statistic, which corrects for chance agreement (Cohen, 1960). The pattern of agreements and disagreements were tested using a two dimensional matrix ('confusion matrix'). Cohen's kappa (κ) statistic was calculated as:

$$\kappa = \frac{\text{Proportion of agreement} - \text{proportion expected by chance}}{1 - \text{proportion expected by chance}}$$

(Cohen, 1960)

A Kappa value of 0.40-0.60 is 'fair'; 0.60-0.75 is 'good' and above 0.75 is 'excellent' (Robson, 1995). The resolution of coding ambiguities or disputes was considered in advance of collecting the data (Cavanagh, 1997). The inter-rater reliability check or reproducibility of the coding of two parental interviews revealed a Kappa value of 0.95 (excellent) for the first interview (agreement on 85 of 91 codes) and 0.97 (excellent) for the second interview (agreement on 111 of 114 codes).

Transferability was addressed by providing a description specifying everything that a reader may need to know to understand the findings, i.e. a 'thick description' (Robson, 1995). An enquiry audit addressed dependability with a documented record of the decision trail and processes kept. Confirmability is achieved when credibility and transferability are established and an outside person can follow the audit trail and establish trustworthiness of the study (Robson, 1995).

Validity of a category or variable infers that there is a relationship between the concept being investigated and the category emerging from the data (Cavanagh, 1997). Three forms of validity assessment can be considered for content analysis: content, hypotheses and predictive validity (Cavanagh, 1997). Content validity was addressed by using an expert (research supervisor) to support category production and coding issues; but this is a weak form of validity as it focuses only on a single variable at a time (Cavanagh, 1997). Hypothesis validity was tested by determining if the content analysis produced results in keeping with theoretical arguments; but this approach has a potential weakness in that if relationships emerge from the data that are contrary to the theoretical foundation of the study or inconclusive, are the variables or hypotheses invalid? (Cavanagh, 1997). Predictive validity was not tested, but refers to the extent to which predictions about happenings are shown to actually occur (Cavanagh, 1997).

As well as a deductive approach of data gathering to test predefined theory, an inductive approach to data analysis was employed (Pope & Mays, 1996). That is, a process of moving from data towards generalisations, hypotheses or theory to generate and interpret themes (Pope & Mays, 1996). Twenty-two themes were identified following second-level coding, plus a further two themes for future analysis ('care received in hospital' and 'care received after PICU discharge'). The definition of each theme and category are displayed (Appendix IV). The index tree number assigned in the NUD*IST database represents the number of each theme. The first index number (1) is not shown as a theme as this represented base data (information on child and parent demographics).

3.3 Results

The findings are summarised by child and parent demographics, and although all aspects of the consultation were interesting, the theme titles (deductively or inductively driven) of child's illness, health and QoL will be summarised only, with quotations from the parental interview transcripts. Twenty-one parents were interviewed; five interviews were conducted in hospital (research site) and sixteen interviews were conducted in the family home. Interviews lasted from 30-60 minutes (mean = 42.1 minutes); all were audiotaped.

3.3.1 Sample representativeness

The time periods for sampling were expanded from 9-15 months and 1-3 months as the previous time periods of twelve months and three months did not provide enough children to be sampled who met the inclusion criteria. Forty-five children were identified from the sampling frame for the 9-15 month group (Feb-Aug 1998) but this number was further reduced owing to one misdiagnosis, three deaths, two duplicate admissions (surviving), ten duplicate admissions (deaths) and one duplicate admission with the other timeframe. Therefore, 28 children were eligible to be sampled. Nineteen parents were randomly selected and invited to participate; thirteen parents agreed to participate, but two parents failed to attend for a hospital-based interview on two and three occasions. A total of eleven parents from the 1998 sample therefore participated in an interview.

Seventeen children were identified in the 1-3 month sampling frame (Feb-April 1999), but this number was further reduced because of one duplicate admission (surviving), one death, one child still in hospital and one child who it was not possible to verify their

current health status. Thirteen children were eligible for sampling and a total sample was therefore recruited. Ten parents agreed and participated in an interview.

3.3.2 Child demographics

The sample comprised eleven male and ten female children, with an age-range of 18-1804 days (mean = 335.2 days). Most children were in the 0-6 months age-range (Table 3.1) on PICU admission. The child's PICU length of stay ranged from 4.0-20.7 days (mean 8.1 days).

Age on PICU admission	Number of children
≤ 6 months	12
7-12 months	4
13-18 months	1
19-24 months	2
> 24 months	2
Total	**21**

Table 3.1 Frequency of child's age on PICU admission (months) – Phase I

Most children were an only child (n=9); fifteen children had 1-4 siblings (Table 3.2).

Sibling history	Number of children
None	9
One	6
Two	2
Three	3
Four	1
Total	**21**

Table 3.2 Child's sibling history (Phase I)

The children were admitted to PICU with varied respiratory diagnoses (Table 3.3); fifteen children were admitted with an acute respiratory illness and six children with an acute respiratory illness and underlying chronic respiratory or other illness.

PICU admission diagnosis	Number of children
Bronchiolitis	7
Pneumonia	5
Respiratory failure	4
Asthma	2
Croup	2
Bacterial tracheitis	1
Total	**21**

Table 3.3 Frequency of PICU admission diagnoses (Phase I)

Ten children had never been admitted to hospital prior to their PICU admission; seven children had been admitted to hospital 1-4 times previously. Sixteen children had never been admitted to a PICU before, one child had been admitted twice previously, and another child had been admitted four times previously.

3.3.3 Parental demographics

Twenty-one mothers were interviewed, with six mothers requesting another carer to participate (five fathers and one step-grandfather). Most of the parents were Caucasian; two were Asian. Most of the 27 parents who were interviewed were married (n=21), two parents were single, three parents were living together and one parent was separated; parental employment status is displayed (Table 3.4).

Employment status	Number of interviewees
Part-time employment	7
Full-time employment	6
Unemployed	4
Housewife	10
Total	**27**

Table 3.4 Parental employment status (Phase I)

Mothers were aged between 20-44 years (mean = 30.3 years); fathers who contributed were aged 20-49 years (mean = 31.2 years). Most parents were aged less than 35 years (Table 3.5).

Age range (years)	Number of interviewees
20-24	7
25-29	5
30-34	7
35-39	4
40-44	3
45-49	1
Total	**27**

Table 3.5 Frequency of parental age-range (Phase I)

3.3.4 Thematic content analysis

The theme titles and categories are summarised in a thematic content analysis matrix (Appendix V). Matrices are useful for bringing together relevant data in a way that will encourage the drawing of conclusions (Robson, 1995). Only a small proportion of the data is usually displayed in matrices, it is therefore crucial that there are explicit decision rules for what is included and that these rules are fully documented (Miles & Huberman, 1984). The number of codes per category, and hence per theme, were identified and frequencies counted utilising the search facilities in NUD*IST (Appendix V). The ranking of themes by frequency of parental response and their overall classification is presented (Table 3.6).

The theme with the highest ranking was 'parental understanding of the child's illness' (585 codes), which reflected Leventhal's self-regulatory model of illness representations. Themes describing health or QoL represented between 51 and 216 codes per theme. The theme of 'negative consequences of child's illness' also ranked highly (339 codes), as did the theme of 'stressors' (388 codes). The themes relating to the child's illness, health and QoL will be presented in more detail.

Theme ranking Order	Theme number	Overall classification	Theme name	Total no. of parental responses or codes
1	5	Illness representation	Parental understanding of child's illness	585
2	3	Other	Stressors – factors causing parents stress during their child's illness	388
3	19	Other	Negative consequences of child's illness	339
4	2	Other	Medical history	286
5	17	Other	Factors affecting parental child health perceptions	216
6	6	Other	Coping with child's illness	215
7	4	Other	Parental wellbeing	208
8	14	Other	Specific health	198
9	18	Other	Positive consequences of child's illness	175
10	21	Other	Neutral consequences of child's illness	118
11	11	Other	Disabilities and quality of life	116
12	12	Other	Factors affecting parental quality of life perceptions	114
13	7	QoL	Global good quality of life	112
14	13	QoL	Global health	109
15	23	Other	Reference to health state pre PICU	97
16	15	QoL	Global unhealthy	92
17	20	Other	No consequences of child's illness	77
18	16	QoL	Specific unhealthy	52
19	8 9	QoL	Specific good quality of life Global poor quality of life	51 51
21	22	Other	Consequences of child's additional illness(es)	14
22	10	QoL	Specific poor quality of life	4

Table 3.6 Ranking of themes by frequency of parental response (Phase I)

3.3.5 Description of themes about a child's illness

3.3.5.1 Inductive, data driven themes

Eight themes arose from categories related to parental descriptions of their experiences on PICU.

Theme 2: Medical history (286 codes)

This theme comprised categories relating to the child's past medical history, including previous hospital admissions.

Theme 3: Stressors – factors causing parents stress during their child's illness (388 codes)

This theme included factors relating to the child's condition (85 codes), ward environment (11 codes), PICU environment (44 codes), level of care delivered by health professionals (64 codes) and relationships including partner (14 codes) and family (40 codes), which

parents described as causing them stress. External stressors were also described such as travel and work (37 codes), and managing their child's illness pre (14 codes) and post (41 codes) PICU admission.

Theme 4: Parental wellbeing (208 codes)

This theme included a category on the expression of emotions, including fear, anxiety and worry (188 codes):

> *"Everything were running through my mind. I thought, I hope he pulls out of it, and all this lot. I were always crying, all time, and then I started crying on the phone, didn't I?"* (Parent 10)

> *"I was just absolutely, erm, I was just really worried wasn't I? I sort of went 'off my tree' at that particular point."* (Parent 15)

Theme 6: Coping with the child's illness (215 codes)

This theme comprised categories on how parents coped with their child's PIC illness, including seeking information and support (12 codes), family support and visiting (83 codes), information sharing by health care professionals (69 codes), coming to terms with the situation (23 codes), and attitudes of health care professionals (16 codes).

Themes 18-21: Consequences of the child's PIC illness (175, 339, 77 &118 codes)

Parents described the consequences of their child's PICU illness in themes of positive (Theme 18, 175 codes), negative (Theme 19, 339 codes), none (Theme 20, 77 codes) or neutral consequences (Theme 21, 118 codes). The themes of positive and negative consequences will be integrated and presented in more detail.

Themes 18 and 19: Positive and negative consequences of the child's PIC illness

Parents described respiratory symptoms that had improved (Theme 18, 19 codes) or deteriorated (Theme 19, 60 codes) following PIC. Parents also described how their child's growth and development was affected after PICU discharge, in terms of their diet. For example, some children had an increased appetite and gained weight (Theme 18, 23 codes), while others were not eating well and required tube feeding (Theme 19, 34 codes). Parents also described varied behavioural characteristics in their children following PICU

discharge. Some children were more outgoing, sociable, strong willed, chatty, or full of energy (Theme 18, 38 codes); others were quieter, sought attention, were disinterested or withdrawn, or did not want to play (Theme 19, 59 codes). Parents described cognitive aspects in their children, such as being more alert on PICU discharge and having no memory of the PICU episode (Theme 18, 24 codes). One parent described how her child was behind at school.

In terms of the child's development, ten parents identified progress (Theme 18, 16 codes):

> *"He's gaining weight, he's started to grip, and he's started to look round, he's started to recognise things."* (Parent 10)

One parent described that her child moved more, another parent said that her child talked more; other parents described their children developing normally. However, according to eleven parents, some children regressed in their development post PICU discharge (Theme 19, 52 codes):

> *"She went straight back into nappies, and she would not say a word to anybody for at least a month..... She'd lost about 2-3 month in development wise...She's ill quite a lot and it puts her back a bit."* (Parent 5)

> *"When she came home she couldn't walk, she couldn't talk, she couldn't feed herself."* (Parent 2)

> *"When he came out* (of PICU) *he couldn't walk, we went down to the children's ward in a chair. It was a couple of days before, physio had to come and get him to start walking again...His potty training ... that went back a bit."* (Parent 25)

> *"Getting the suckling reflex back again, that was a bit of a long slog, because she'd had a sore throat with the tube being in so long, and erm, the coughing had disrupted her, and I think she'd just basically forgotten how to suck. She was tube fed for nearly five weeks."* (Parent 22)

"The weeks that he spent in intensive care did set him back quite a lot 'cos he lost sort of all, all that he'd learnt in the last five months." (Parent 11)

Two parents described improvements with their child's sleep patterns following PICU discharge (Theme 18, 4 codes):

"He sleeps brilliant on a night now, like I say, six to six, twelve hours, before he was an awful sleeper on the nights." (Parent 25)

"She was sleeping with us you see before she went into hospital. When she went into hospital she had to sleep on her own. She had a lovely big crib (at home) *and she just hated it, she hated being on her own. So when we brought her home you see, put her in her cot, out like a light, she settled no problem. So that was a positive thing actually."* (Parent 22)

However, nine parents described sleeping difficulties in their children (Theme 19, 29 codes):

"When she first came off intensive care, she was awake for two and a half days, she never slept day or night, and then she did eventually drop asleep...She used to have nightmares or whether she used to see things, um, she used to hallucinate, she used to try and grab things that weren't there, that only she could see...She did sleep a lot more when she came home. She seemed tired, but eventually she's got back into a normal routine." (Parent 2)

"She was throwing herself round the cot and squealing and she wouldn't be comforted and she couldn't sleep, and oh, that was awful." (Parent 12)

"She was sleeping through before we went into hospital and then from coming out she's never slept a night." (Parent 24)

"When she first came out of hospital, it was a couple of days before her first birthday she come out. She was in a cot then, it was a cot/bed, and she just screamed and screamed. She just would not go in this cot because of the bars.

Because when she was in intensive, before she was in (name of local hospital), *she were in a cot then. Um, I don't know if she remembered, or, but she was really petrified. She just would not sleep; she had to sleep on couch. And then err, I took the side of the bars off and she was fine, when she was like in a bed, from when she were one year's old."* (Parent 5)

Eleven parents described emotional characteristics in their children post PICU discharge, such as being happy and content, smiling more, giggling, or having a closer emotional bond with them (Theme 18, 25 codes). However, fourteen parents described their child as being unhappy, crying, wanting cuddles and being clingy, screaming or getting easily upset or frightened (Theme 19, 48 codes):

"When she came home, um, I mean she's got a doll, a teletubbie, a La-La doll, and when you press it, it speaks. And even to this day she won't entertain it; it frightened her when she came home. And now, if you show her it, if you show her it now, she just "no, put it away, don't want it, don't like it". Um, she has a clown as well, which, um, helps you tie your shoelaces and buttons and stuff like that. She only has to see foot in cupboard and she's "shut door, shut door", she don't like it. Um, so things that frightened her then are stuck with her. She still won't entertain it." (Parent 2)

"If anybody came, I always remember, we had people coming, we had a kitchen fitted and all this sort of thing, and when people came in, she used to run and hide. Um, or she'd say, "man's coming..No, no, no." You know, and she'd come and sit on your knee, err, but eventually that's stopped now, she's not bothered anymore when people come." (Parent 2)

"I mean his feet, he didn't like being, you couldn't go near his feet at one point, even just to put his socks on. He cried, he'd had that many stabs on them." (Parent 21)

One parent described how their daily routine was unchanged following PICU discharge or changed very little. Eight and twelve parents identified positive and negative aspects of health respectively (Theme 18, 12 codes; Theme 19, 19 codes respectively). Parents

described survival as being a positive aspect of their child's health (Theme 18, 12 codes), while negative aspects included the child being generally weak, being more prone to illness, having further chest infections or asthma attacks (Theme 19, 19 codes).

Parents described how their relationship with their partner was affected during the PICU episode and subsequently. Eight parents described negative aspects including a strain on their relationship with their partner during the PICU episode (Theme 19, 28 codes). Nine parents described positive aspects including a strengthening of their relationship with their partner, and becoming closer (Theme 18, 14 codes). Some mothers described how they felt more confident about their child's illness and had increased knowledge in recognising signs of respiratory distress:

> *"I know when his chest is bad. I know he's fine because his colour is all right. The only time I ever worry is when his colour goes off 'cos I know then that he's having problems."* (Parent 11)

Some mothers described how their relationship with their partner or other children was affected by their child's illness. One mother, who had a baby with chronic lung disease on low-flow oxygen at home stated:

> *"We can't have any time to go out together and it's putting a strain on us. And plus, my little girl, I mean, she loves him* (child) *to bits and she hasn't got a problem with her dad, she's got a problem with me. I think she feels jealous I'm with him all the time 'cos, I mean, she's been with us on her own for five years... I mean I love her and I have a bond with her, but I think it's because I have a stronger bond with him, you know, erm, I'm protective against him. I'm scared of people looking after him in case anything happens and we're not there and then you now, I feel it will be my fault, you know."* (Parent 13)

One parent described feelings of worry:

> *"I'm a lot more paranoid than I was, any slightest illness and I'm like straight to the doctors and that. I mean, 'cos about four month ago, she had a really bad chest, and I was really, really scared that it was croup coming back again.*

So, I kept taking her to doctors everyday, you know proper neurotic mother and all that." (Parent 5)

"I'm very watchful. I mean, I think, God it's happened once, can it happen again?" (Parent 26)

"Since he's been on intensive care, I love him very much when he came back. I can't bear to see him cry or anything, it's just like no good." (Parent 6)

Another parent whose child was hospitalised for most of his life, and then was discharged home commented:

> *"Our whole life has changed since we've got him home, yeah, obviously, erm, it's totally different now to what we were before, you've got to think about what you're doing, where you're going. You've got, err, with his oxygen requirement you think about where you're going as well. You couldn't just go for a meal in a restaurant, and I wouldn't take him in a smoky atmosphere for instance."* (Parent 21)

3.3.5.2 Deductive, theoretically driven themes

Theme 5: Parental understanding of child's illness

The theme of 'parental understanding of child's illness' was driven by health psychology theory on illness representations and cognitions. The theme reflected Leventhal's model of illness representations, with parents describing their perceptions of the cause of their child's illness, timeline factors, and cure and control factors and symptoms (585 codes).

Nineteen parents described respiratory symptoms of their child's PIC illness, including wheeziness, general colour, cough, blocked nose or snuffles, nocturnal cough or breathlessness (Theme 5, 104 codes):

> *"...she just stopped breathing... her chest, her throat were constricting and it wouldn't let her breathe.... she were going blue round lips..."* (Parent 5)

> *"I noticed she'd started coughing during the night, when I fed her during the night, she started coughing really bad, and you know sometimes she wasn't getting her breath."* (Parent 24)

Parents described how they sought advice from friends (7 codes), family (9 codes) or health care professionals (56 codes) to help them to understand their child's illness. Parents also described their perceptions of the cause of their child's illness such as a virus, infection from birth, or immune suppression (59 codes), and the duration of their child's symptoms (27 codes) and time it took for their child to return to normal (37 codes). Thirteen parents described cure and control consequences of their child's PIC illness (29 codes):

> *"He got it* (illness) *once, um, were it after intensive care? Um, I think we'd been shopping, and we got caught in rain, and he was bad after that, so we just, if it's spitting or 'owt, he comes in you know, we don't have him out in the rain."* (Parent 7)

> *"In the winter time, the only time she suffers with her asthma, if she gets a cold and I can't get the cold first. As soon as it goes on her chest the asthma starts. So, if I can sort of knock the cold back before it goes to her chest she's fine."* (Parent 26)

3.3.6 Description of themes about QoL

3.3.6.1 Inductive, data driven themes

Eight themes on health and QoL were identified and included global and specific themes with poor or good aspects (Themes 7-10, 13-16). Global themes were defined as parental perceptions of a child's health or QoL in general terms. Specific themes were defined as parental perceptions of their child's own health or QoL. Themes were also identified from categories describing factors affecting parental health or QoL perceptions (Themes 12 & 17). These themes were primarily data driven but also reflected theoretically derived dimensions of health described in other child HRQoL measures. Each of these themes will be discussed in turn.

Theme 7: Global good QoL (112 codes)

Parents described emotional, behavioural, cognitive, family, environmental, social, health and physical (nutritional) aspects in respect of a child with a good QoL. These aspects are also described in the literature on child HRQoL measures. Twelve parents perceived a child's QoL to be good if the child was happy, loved and received attention, particularly from their parents (25 codes):

> *"I think affection is probably the main one for quality of life"* (Parent 22)

> "*They* (children) *always need to know there is someone there for them."* (Parent 12)

> *"Most important for a child is love and attention."* (Parent 11)

Four parents described the importance of a child's behaviour in terms of good quality of life (4 codes):

> *"...fetch 'em up so they don't get in trouble with police."* (Parent 10)

> *"..they* (children) *need to be shown what's right and wrong, what's good and bad.."* (Parent 25)

The importance of education and learning (cognition) was also described as an important aspect of a child's QoL by five parents (13 codes):

> *"They* (children) *learn from you, if you're that close, you can learn from each other, like a two-way thing."* (Parent 4)

> *"...showing them new things, giving them new things to discover."* (Parent 4)

> *" .. a good quality of life – to be brought up to know the rights from wrongs, to have a good education.."* (Parent 26)

Thirteen parents described the importance of the parental role and family support in determining a child's QoL (21 codes):

> *"Being in a family, yeah, a family. I think it's important to, erm, to have two parents there. It must be really hard if you're a single parent for whatever reason. It definitely helps to have the influence of at least two adults.....They* (children) *always need to know there is someone there for them."* (Parent 12)

> *"I think your family is one of the most important things. I mean, erm, I suppose even if you haven't got any money, at least if you've got your family it's you know, they can give you support... You need a family, you don't need anything else. As long as you've got a family you know, you know you're right 'cos you've got somebody to turn to."* (Parent 26)

> *"..having parents that love 'em, more than anything."* (Parent 5)

Five parents described the importance of providing food for children and keeping them clean and well nourished (9 codes), and eleven parents described providing a warm place to live to ensure a child has a good QoL (20 codes):

> *"I mean first and foremost the* (children) *need to be fed and clothed and have heating...This is going to sound awful snobby, and I don't intend it, but I think they* (children) *need a good home, a good place to live."* (Parent 12)

> *"..they've got a home, so they're warm and comfortable."* (Parent 22)

Seven parents described the importance of social interaction for the child with other children and adults, and the value of play (13 codes):

> *"I think that socially children, especially when they're getting erm, over a year, need to mix... they need to be able to socially develop I think."* (Parent 12)

"..you need to talk to 'em, you need to read to 'em, you need to like play with them." (Parent 11)

"That they're like playing out and stuff, because I think if they're stuck in the house they seem more miserable. They should be like let to run everywhere and play a lot..." (Parent 3)

Six parents identified *"having decent health"* as an important aspect to a child's QoL (7 codes). One parent stated:

> *"If they're healthy, they've got to have a good quality of life, haven't they?"*(Parent 19)

Parent 10 described the importance of a child having regular checks with the health visitor and the school doctor.

Theme 8: Specific good QoL (51 codes)

Parents described their own children as having a good QoL in relation to several factors. Five parents described inter-personal interactions (9 codes), three parents described behaviour (4 codes), seven parents described family support (16 codes), four parents described emotions (4 codes) and two parents described diet (2 codes). For example:

> *"I think he's got a good quality of life. I mean we both love him and we, you know, we're there for him."* (Parent 13)

> *"He has to eat his food mashed up, but I don't think that makes his quality of life bad. He eats the same as us, it's just mashed up."* (Parent 18)

> *"He has asthma, I wouldn't say that stops him from doing things, so, therefore he has a good quality of life."* (Parent 25)

Theme 9: Global poor QoL (51 codes)

Four parents described a child with a poor QoL as an unhappy child, and one who was alone (6 codes). Parent 20 described children with a poor QoL as:

"Receiving no love or attention from their parents...If the parents don't love their children, you know they're obviously not going to buy them things that they probably need for development of things."

Parent 5 reiterated this comment:

"They've got everything that they need, but the parents aren't there for them. You know, they've got the top shoes and the top cars, but when they (children) *really need someone, there's nobody there."*

Parent 11 described a child with a poor QoL as:

"Someone that isn't loved. Somebody that like their parents don't do 'owt with 'em. They don't, not so I mean I suppose they still might feed 'em properly, but they don't talk to 'em, they don't play with 'em and they don't encourage them to do anything. They're just there and they just leave 'em. Not necessarily neglect 'em, but think, you know, that by, you know like, as long as they've got toys and as along as they've got games and as long as they've got telly then they're fine. You need to talk to them."

Many of the parents described a child with a poor QoL as being the opposite of a child with a good QoL:

"Basically the opposite of good quality of life, just like lack of love and attention from parents". (Parent 20)

Some parents discussed the role of money and material things in relation to a child's QoL:

"I think sometimes you can actually give too much materialistic things without giving time. You don't have to spend a lot of money on a child to, erm, give time. You can spend a lot of money on a child, on children, bringing them toys, erm, and various material things, but it's no good spending the money on things if you're not prepared to the give the time to actually show them (children) *how to utilise their toys."* (Parent 15)

Parent 19 said that a child may have luxuries such as toys and TV, but would not have a good QoL if they had these luxuries but were also being abused.

Six parents described the child's environment as affecting a child's QoL (9 codes) and two parents described health status as an influencing factor (4 codes):

> *"A lot of these kids in here* (hospital) *just don't experience things, play, because they just don't, they're laid in a cot or they're just laid in, even worse, on ventilators and things for months and months, which that to me is not a quality of life."* (Parent 21)

> *"If they* (children) *come from a background that's socially deprived, I suppose, erm, living in a run down home.... And there's ten of them in one room and you know they can't afford to feed them properly and this kind of thing, or clothe them properly, or, erm...perhaps the parents are heavy smokers and things, which I think is quite a major thing. I can't abide people that smoke around small children especially when they've had, you know, respiratory illnesses I think a lot of that makes, err, has an effect on the child's health."* (Parent 12)

Parent 10 said that a child with a poor QoL was neglected by *"not being fetched up in a good atmosphere or environment."* Two parents mentioned a lack of *"proper food"* or *"poor diet"*.

A child's health was also related to poor QoL, but not necessarily poor health indicating a poor QoL. Parent 12 stated:

> *"You have to have the physical things in life* (being fed) *first and foremost because if you haven't got those, you know, then they're* (children) *gonna be ill."*

Theme 10: Specific poor QoL (4 codes)

Two parents perceived their own child's QoL to be poor in relation to inter-personal interactions such as not being able to communicate (3 codes), and one parent described poor QoL in relation to their child's behaviour such as tantrums (1 code).

One parent believed her child's QoL was quite bad:

> *"I don't feel that it is bad, it's quite bad but err... With her, err, um, like not understanding to communicate to us."* (Parent 1)

Theme 11: Disabilities and QoL (116 codes)

Parents described factors affecting their perceptions of the QoL of a disabled child (72 codes) and their assessment of the QoL of disabled children (44 codes):

> *"If you lived there as a 'vegetable' to my mind you don't have a quality of life. If someone is born physically disabled from birth they don't know any different, so to them it's, you know, how they are and they accept that and they get on with it and they would enjoy life better. Whereas, someone who had, say an accident in their teens and was used to running round playing football or things like that, they would feel their quality of life is poor."* (Parent 21)

Theme 12: Factors affecting parental QoL perceptions (114 codes)

Several factors influenced parental perceptions of a child's QoL, including parental experiences of children, parental beliefs and values, and parental understanding of the concept of QoL.

Nineteen parents discussed children with disabilities in relation to QoL (44 codes) and twenty parents stated their beliefs and values (89 codes):

> *"I don't suppose it matters what's wrong anywhere down the line, you can still make their* (disabled children) *quality of life as best as you can, can't you?"* (Parent 24)

"Everybody's an equal, and if they (children) *did have a disability, you'd work with that and overcome it. Well, not overcome it, but you'd make, you'd, you'd sort of lie, um, how can I put it? You'd, you'd cater for that and you'd just carry on as normal as you could."* (Parent 4)

"If I had a normal child, I don't know what I would be saying then." (Parent 1)

"She could have come home with any, any disability, it didn't matter, I just wanted her home... anything would have been better than not having her... but had she been born with that way (with a disability) *it might have been different."* (Parent 2)

Other parental beliefs included allowing children to develop socially, views on material things, working and providing the best for a child:

"If children are not given the chance (to socially develop) *then I think that's quite bad for them mentally, mental development that way, 'cos if they can't mix they can't learn, they can't learn what's right and wrong and that kind of thing. So, I think that's quite important."* (Parent 12)

"You don't need to have loads of money for 'em (children) *to know how they've got a quality of life and that they're loved."* (Parent 11)

"The children over the road are limited in the amount of things that they can do because of the money and the resources she (mother) *has. All their clothes they have are pass me downs, whereas my children get their own clothes, their own you know, they're a lot more independent."* (Parent 9)

"We, nobody's got a lot of money or anything, but as long as you're there providing what they (children) *do need emotionally, then everything else will take care of itself really."* (Parent 5)

"There's financial, that's only why I work full-time, you know, you don't get anything in life if you don't work for it to a certain degree. I mean a lot of

people say you miss out on children, you know, when you work full-time. But I also think you give them a better, a better you." (Parent 9)

"I mean, just 'cos you're a working mum, it doesn't mean to say that you can't look after your children because you know, it's just the same quality times, even if it's only half an hour a day, you know, if you're spending quality time with them." (Parent 22)

"You can never give a child too much love and affection you know." (Parent 22)

Parents described how their values had changed since their child's illness:

"Before, I'd been on half pay maternity leave, you worry about paying the mortgage and everything, and all that had just gone out the window. It was just like well it's not really important you know, it's, it's just material things, and it's not really important you know." (Parent 24)

3.3.7 Description of themes about health

3.3.7.1 Inductive, data driven themes

Themes on child health were data driven, but reflected theory on child health.

Theme 13: Global healthy (109 codes)

Eleven parents described emotional characteristics of a healthy child as one who is loved, happy, smiling, laughing and joking or content (22 codes):

"If a child is happy, then they're healthy, aren't they?" (Parent 20)

Ten parents also described behavioural aspects, such as a child who has no tantrums, is full of energy, has a pleasant manner, or is bubbly (18 codes):

"Happy, smiley, loads of energy, wanting to play, generally running about and making a nuisance of theirselves." (Parent 2)

Cognitive aspects of a healthy child also featured in descriptions from five parents (6 codes):

> *"I think they've got to be mentally OK as well... to be fully healthy."* (Parent 21)

> *"I think his mental state of being as well as his inner state of being, i.e. his chest and everything. If they're mentally happy, and mentally stable, the rest of it, you always get the illnesses, but they seem to cope with it a lot better, you know what I mean?* (Parent 8)

> *"I mean a happy child is a child that learns and develops. A child that can learn and develop without any injuries or illnesses, because it's not hindering them."* (Parent 9)

Seven parents also described the importance of inter-personal interaction and play to health (10 codes):

> *"When they're actively involved, they're socialising with other children, they're laughing, they're smiling... they're participating with each other and with adults generally."* (Parent 15)

Aspects of a child's growth and development were also stated by thirteen parents as important to health (24 codes) and ten parents described the importance of a balanced diet, good appetite and the child being well fed (16 codes):

> *"Able to walk about and to communicate, like speaking and that"* (Parent 1)

> *"Erm, I think physically, it's obviously if they're the right sort of height and weight for their age group."* (Parent 12)

> *"I think developing as they should, I think they're most important bits"* (Parent 4)

"You know as long as they're eating and drinking and sleeping when they should be and running about and whatever." (Parent 11)

Two parents described a healthy child by its appearance in terms of its complexion, colour and not *"looking poorly"*(9 codes). Family and environmental aspects were also each mentioned by one parent (1 code each).

Theme 14: Specific healthy (198 codes)

Parents related their experiences of health to their own children by describing aspects of health that were important to their child. For example, six parents described emotional aspects (12 codes), three parents described inter-personal interaction (3 codes), five parents identified dietary aspects (7 codes), six parents identified growth and development factors (12 codes), three parents identified behavioural aspects (4 codes), and one parent identified the child's appearance (1 code):

"She's happy, she's always laughing, she eats well... she's always laughing and playing."(Parent 20)

"You know even though all that has happened, she is generally a healthy child. She eats well, erm, you know, she has a normal, you know she has a normal habit of what children do at that age, you know, she's not, erm, there's no areas of her progress that's less than..." (Parent 24)

"I'd say she (sibling) *was healthy, more healthy than what he* (child) *is because she wasn't born as early as him."* (Parent 13)

"She's happy, she's quite robust. She gets on with everything, it's just, you know, she does what she wants to do." (Parent 9)

Twenty parents also described their perceptions of their child's own health before PICU admission (71 codes):

"His health is really not that good... mostly he's been poorly." (Parent 6)

"She was a fat little thing, erm, I would have said that she looked really, you know, absolutely full of health." (Parent 12)

"I mean that's really the only thing that's ever been a problem, his chest. I mean the rest of his health, you know, apart from that, he's in good health." (Parent 11)

Nineteen parents described their perceptions of their child's health post PICU discharge (88 codes):

"He's got a lot better, and now he's, he's pretty healthy. I mean he gets out of breath 'cos of like being sick, he's just, that's it, that's how much it takes to wear him out." (Parent 14)

"You can tell he is healthy, he has much more energy, he's smiling." (Parent 21)

"Whether it's due to ICU or not I've no idea, erm, we've had a very bad time with him...He was fine for about six weeks after he was discharged from (name of local hospital)... *and then he got a huge lump on his neck."* (Parent 15)

Theme 15: Global unhealthy (92 codes)

Parents identified the characteristics of an unhealthy child; most of these characteristics were opposite to those described for a healthy child. Eight parents described a child who is unhappy or sad or receives no love (13 codes), three parents described an unhealthy child as having dirty clothes or living in an unclean home or one who has no opportunities in life (6 codes). Fifteen parents identified an unhealthy child as being withdrawn or introverted or does not play or is unsociable or has no energy or sleeps a lot (37 codes), three parents stated the child has a physical disease or is poorly (4 codes). Ten parents said the child does not eat or is not well fed (15 codes), four parents stated the child does not interact or talk to others (5 codes), and six parents described the appearance of an unhealthy child (12 codes):

"Usually when a child gets poorly it's crying and ratty." (Parent 22)

"Not getting well fed, always wearing same clothes, not seeing GP all time, not getting all love and attentions." (Parent 10)

"Maybe its home circumstances weren't, erm, like as good as what they could be really...maybe you know, if it's not very clean." (Parent 24)

"(An unhealthy child is) *quiet and withdrawn."* (Parent 19)

"Looks pale and withdrawn... one that just sits there, doesn't talk, doesn't interact." (Parent 5)

"An unhealthy child, to look at an unhealthy child, would be, err, quiet, shy and withdrawn." (Parent 8)

"Pale, thin, not doing now't, crying..." (Parent 4)

"One who's off his food, lethargic, doesn't want to do anything, might have a temperature, a cold." (Parent 17)

"By the way they look, I mean like their skin, I always tend to go by their skin...I mean, usually you can tell, I mean if they've flaky skinned, or pale, or blotchy.... that's what I look at, yeah, their skin and their eyes." (Parent 22)

Theme 16: Specific unhealthy (52 codes)

Parents expressed characteristics of an unhealthy child in relation to their own children. Four parents described emotional characteristics (6 codes), six parents stated behavioural aspects (10 codes), two parents stated dietary factors (2 codes), and one parent described their child's growth and development (1 code):

"How he were when he stopped eating. You could see in his face how it changed, from him having like a littler grin, just seemed right serious, and if he smiled it would crack his face. He just couldn't move his face." (Parent 3)

"I think temperament would be the first thing, 'cos she's usually, if she's crying you usually dance round the room, you can do something stupid and, or show her one of her toys, and she's laughing again. But if she didn't and it went on and on, I'd think now there's something not quite right." (Parent 22)

"I think he is not healthy because he was born early..and he's on all this (points to oxygen), *to me he's unhealthy."* (Parent 13)

"It's just the fact that she has to have inhalers, it is drugs." (Parent 9)

Eleven parents also reported symptoms of illness, such as a temperature, cough or cold, sore throat, pain, pallor, lethargy, flushed face, runny nose, or their child not eating well (30 codes) and two parents described treatment factors (3 codes).

Theme 17: Factors affecting parental health perceptions (216 codes)

Several categories emerged from the analysis relating to the theme of factors affecting parental perceptions of health. Although these categories were data driven, they reflect psychological theory of illness perceptions as described previously. Seventeen parents identified their experiences of healthy children (51 codes), eighteen parents described their own values and beliefs (55 codes), ten parents described parental intuition (28 codes), nine parents identified their experiences of ill health (13 codes), seventeen parents stated their understanding of the concept of health (55 codes), and eight parents described their attitudes towards health (14 codes).

The following quotations relate to parental experiences of healthy children:

"Whether it's, that's the way children are, only ever having had her and nothing to compare it with I don't know." (Parent 2)

"With him being the first, I don't know you know, what he should be doing as well as somebody who's got a couple of kids...I look in books and what have you." (Parent 4)

"I think if you've already got children, older children, you know what they should be doing by what age. And you know the sort of limits of you know, when they're early and when they're late, and you know they get that they've got to sort of be able to start doing things." (Parent 12)

"I've never like seen a healthy child. I mean, I wouldn't say like I've lived with a healthy child, because obviously (child's name) *is my first one. It's hard for me to compare, and I don't have any younger brother and sister that I can remember of how they grew up and that."* (Parent 1)

The following quotations relate to parental intuition:

"You can just tell when they're not feeling themselves you know" (Parent 1)

This quotation is interesting as it is the same parent who said that she had no experience of children (see above).

"I knew that he wasn't right." (Parent 15)

The next quotations illustrate parental understanding of health:

"I think especially with, you know, with being a nurse, you have a bit of, you know they say a bit of knowledge is dangerous and it is, isn't it?" (Parent 24)

"They've (siblings) *have never suffered like* (child's name) *with asthma, bronchitis. Alright, they have their colds, their sore throats, the cuts, the abrasions, err, to me that is a healthy child."* (Parent 26)

"Healthy? It means they haven't been ill." (Parent 6)

"I suppose if they haven't got a disease." (Parent 24)

"I don't think you can single out one thing (important aspect of health). *So, I think if you take, if you start taking things out it, erm, I mean you've got*

to...there's got to be sort of communication there, there's got to be the sort of feeling it's the whole thing, the whole concept of the child's wellbeing, that's the thing rather than an actual single thing." (Parent 15)

"A child with illness, he's got an illness, yes he can be healthy one day, err, he can, you know no pain, anything there, but as soon as he's got pain it's not a healthy child." (Parent 26)

> *"There's like the illness that you can recognise, like, like her, or the sickness or whatever. And then there's like a disease you know, like a genetic disease like* (person's name) *has, and unless he has one of his bad days, you'd never know that he was poorly...Unless you can see it, I don't think you really know."* (Parent 22)

Parental attitudes to health were reported particularly in relation to children with disabilities:

"Look at people with disabilities, and see that they're not healthy. But then again, people, children with these disabilities, you can always see again with their mental frame of mind, their features, whether they're happy or not." (Parent 8)

"I mean you can be healthy I suppose and then have mental problems." (Parent 21)

"A disabled child needs you for the rest of their life, you know." (Parent 13)

3.4 Discussion

This study aimed to describe parental beliefs about health, QoL and illness. Themes were inductively and deductively driven, with the results of the thematic content analysis providing quantitative data to illustrate the most relevant themes to aid item-generation for the HRQoL measure.

Important dimensions of health and QoL, as perceived by parents, were identified in global and specific terms, including behavioural, emotional, general health and illness perceptions, physical, psychological, social, and respiratory-specific consequences post PICU discharge (Grange & Russell, 2000). Respiratory-specific consequences included the type and severity of respiratory symptoms, including the presence of a wheeze, breathlessness, cough, runny nose or snuffles. Other consequences included the impact of the child's illness and respiratory symptoms on the child's daily activities and daily family life. Some parents reported their child's daily activities returning back to normal fairly quickly after PICU discharge, but progress was reported as slow for other children, particularly those children discharged from PICU with an acute exacerbation of an underlying chronic respiratory illness. The dimensions of health described by parents were influenced by a number of factors including parental perceptions of health and illness.

Parents described their beliefs about their child's illness in terms of cause, timeline, cure or control and consequences factors. These factors are reflected in a self-regulatory model of illness representations (Leventhal et al, 1980; Leventhal et al, 1984; Leventhal & Nerenz, 1985). A number of factors were described by parents that affected their perceptions of a child's health or illness, including their own experience of healthy or ill children, their own values and beliefs, attitudes to health and understanding of the concept of health and ill-health. These influencing factors are also described in the health psychology literature (Froberg & Kane, 1989; Schor, Lerner & Malspeis, 1995; Weinman et al, 1996; Levi & Drotar, 1998). Evidence suggests that medical knowledge and/or experience with illness may influence raters' valuations of health states, and that the measurement of HRQoL in children is influenced by child and parent health and illness cognitions, age, cognitive development, sex, parental social status and non-specific effects (placebo) (Froberg & Kane, 1989). The theme of parental wellbeing also featured strongly in the analysis, with parents describing their emotions and feelings, particularly in terms of anxiety during their child's PIC illness. Parental anxiety and adjustment levels are reported to affect parental reports of a child's HRQoL (Levi & Drotar, 1998). Parents described family support as an important area in helping them to cope with their child's PIC illness; receiving appropriate information and support from health care professionals also rated highly. Parental illness perceptions and anxiety can influence HRQoL perceptions.

Parents described the most important characteristics of a healthy child as being happy and content. Social interaction was also expressed as an important characteristic of a healthy child, with the child being able to interact with others and play. Parents also reported psychological and cognitive aspects of health, with the importance of learning and education emphasised. The dimensions of health identified in this study were also described in the systematic review of child HRQoL measures (Chapter 2). These dimensions of health encompass the definition of health provided by the WHO, including physical, mental and social components (WHO, 1948).

The dimensions of HRQoL identified in the systematic review were also noted in the thematic content analysis, namely, disease state and physical symptoms, functional status, psychological functioning, and social functioning (Speith & Harris, 1996). Parents described the disease state of their children pre and post PICU admission, and respiratory-specific symptoms. Parents also described the impact of their child's PIC illness on daily activities. The child's functional status was also described in terms of their health and QoL, and included psychological and social aspects of functioning. Some parents identified the concept of health to include the presence of a disease or illness, which differs from the WHO definition of health. Parental perceptions of QoL differed from health in terms of the importance of a child belonging in a family and the role of parents in providing care and giving the child love and attention. Physical aspects were often reported by parents as important for QoL, including ensuring the child was warm, clothed, well fed and had a home. Emotional aspects featured prominently when parents described children with a poor QoL in terms of those children not receiving love or attention from their parents. The role of the family was particularly emphasised by parents in both themes of poor and good QoL. The family can have a significant impact on a child's health through genetic and familial predispositions, learned health beliefs, and shared physical, social and emotional environments (Schor et al, 1995).

Findings from this study identified the important dimensions of health and QoL in which to develop parent-generated items for the HRQoL measure: physical, emotional, social, psychological, and cognitive aspects. Respiratory-specific components such as frequency and severity of symptoms were also identified. The importance of parental illness beliefs and anxiety levels were also recognised as important components to include within the HRQoL measure as they can influence parental perceptions of HRQoL. Parents placed

more emphasis on the emotional and social dimensions of health, rather than the physical dimensions of health.

The strengths of this study were that face-to-face interviews with parents provided rich and illuminating data on the concepts of child health, HRQoL, and illness perceptions. Areas of uncertainty and ambiguity were clarified with parents avoiding misinterpretations (Cormack, 1991). The research was also rigorously carried out. The inductive approach applied in this study allowed the building of abstractions, concepts, hypotheses and theories from the data (Creswell, 1994). Much of the data analysis in this study was driven by the data itself, but a deductive approach was also utilised to inform the inductive approach to the data analysis. This may be considered a particular strength as the theory of illness perceptions, WHO definition of health, and dimensions of health identified from the literature informed the coding framework for analysis.

It is argued that both predetermined and emergent categories are used in content analysis (Moseley, Mead & Murphy, 1997). Moseley et al (1997) state that it is futile to pretend that a data set is approached with an empty mind, and without ever having read on the research subject; however, an approach which had no flexibility and which offered no opportunities for sudden insights to emerge and be tested, would be equally undesirable. A combination of deductive and inductive approaches to data analysis is thus recommended (Cavanagh, 1997). Content analysis involves the placement of responses or fragments of responses in categories developed in the context of the research, and is claimed to be a research technique for making replicable and valid inferences from data to their context (Moseley et al, 1997). Some argue that content analysis is too simplistic and may fail to produce rich data (Field & Morse, 1985; Cavanagh, 1997), while others argue that counting is indispensable (Miles & Huberman, 1984).

The rigour of the research was also strengthened via the inter-coder reliability checks using Kappa statistics, which revealed findings in the 'excellent' range of agreement. Content validity of the classification system was tested through the use of an expert (research supervisor) to support category production and coding issues. The content analysis produced results in keeping with theoretical arguments concerning parental illness perceptions and definitions of health, thus supporting hypothesis validity (Cavanagh, 1997).

The weaknesses of this study were that the parental interviews were conducted after the child's PICU episode and some parents therefore had to recall events up to fifteen months previously; recall bias was therefore a potential problem. Respondent bias may also have been a problem as respondents generally try to tell the interviewer what they think the researcher wants to hear, and respondents may consistently agree or disagree with the questions (acquiescent response set) (Brink, 1991; Stern, 1991). Respondent bias was minimised by constructing questions in such a way as to avoid a correct answer, and agreement/disagreement response modes. The intention was to elicit what respondents believed to be the correct social response. Interviewing is a specialist skill and requires practice. Conducting further parental interviews with parents may have strengthened the author's interviewing skills. The reliability of the data analysis was addressed via inter-rater coding checks; most parental interviews (n=19) were coded by a single-coder. Thus, indicating that stability may be the weakest form of reliability. Human fatigue, personal bias and a change in perception over time may affect the reliability of the data analysis (Cavanagh, 1997).

The sample comprised mainly Caucasian parents with only two Asian parents interviewed. The sampling criteria of English-literate parents may have precluded the participation of Asian parents in the study. Over-sampling for parents from the ethnic minorities may have improved the Asian representation of the sample. A random sampling technique was used to ensure that each member of the population had an equal chance of being selected. However, a purposive sample, where the researcher uses his or her judgement, based upon the best available evidence, to ensure that the sample selected possesses the characteristics needed for the study, that is 'represented' in the population on some trait or variable may have been preferential (Brink, 1991). Involving an interpreter may also have increased the response rate from parents from the ethnic minorities, but was beyond the financial constraints of the study.

Future directions of this research may include further exploration of the concepts of illness representations and beliefs and the impact these concepts may have on parental coping mechanisms in a PIC setting. However, this is not in the scope of the HRQoL measure or this thesis.

3.5 Summary

In summary, qualitative parental interviews conducted in this study (Phase I) established global and specific parental perceptions of child health and HRQoL, and identified parental illness perceptions. The consequences of the child's PICU admission were also reported and respiratory-specific consequences identified. Bias was acknowledged and measures to reduce these described. Important dimensions of health and QoL, as perceived by parents, were identified; they compare to those dimensions of health described in other paediatric HRQoL studies, namely physical, social, emotional and cognitive functioning (Chapter 2). Face and content validity of the HRQoL measure was supported by involving parents in the development of items and considering their views on the concepts of health and HRQoL.

CHAPTER 4 – Qualitative study to generate items to measure the clinician perspective of HRQoL

4.0 Introduction

This chapter describes a qualitative study conducted in two phases (Phase 1 and Phase 2) that identifies clinicians' perspective of HRQoL outcomes and objective respiratory outcome measures for children aged 0-5 years admitted to PICU with a respiratory illness. It builds upon previous studies conducted in Chapters 2 and 3, which identified points from the HRQoL literature and parental concerns about health and HRQoL. Themes identified from the analysis informed the development of clinician-generated items for inclusion in the HRQoL measure.

4.1 Background

From preceding chapters, the need to ascertain health care professionals' beliefs about HRQoL was emphasised to inform the development of an HRQoL measure. Little prior evidence exists of others' beliefs about HRQoL. Many studies in PIC relied upon the measurement of health outcomes as the 5 D's: death, disease, disability, discomfort or dissatisfaction (Lohr, 1988). Some studies reported functional or psychological and emotional outcomes; few reported QoL and HRQoL outcomes following PIC. It is unclear what the perceptions of health care professionals are regarding a child's HRQoL.

Increasingly there is a movement towards measuring more positive aspects of health outcome, including subjective outcome measures such as wellbeing and QoL, rather than relying on the more negative aspects of health outcome, such as mortality, or objective outcome indicators (Jenkinson, 1994; Bowling, 1997b). Few objective respiratory outcome measures are reported in the literature for use in children under the age of five years (Chowienczyk et al, 1991; Phagoo, Wilson & Silverman, 1995 & 1996; Bridge, Lee & Silverman, 1996). Those that are reported are not used routinely in practice. Subjective measures of HRQoL outcome can complement objective measures, but need to be useful in the clinical setting, practical, easy to use and interpret; they must also be acceptable to patients and health care professionals (Greenhalgh et al, 1998).

This chapter aims to identify the factors that health care professionals or clinicians consider to be important in informing the development of an HRQoL measure for children, under the age of five years, admitted to a PICU with a respiratory illness.

4.1.1 Research aim

- To develop clinician-generated items for incorporation into the HRQoL measure

4.1.2 Research objectives

- What is the clinician perspective of HRQoL outcome assessment for children under the age of five years admitted to PICU with a respiratory illness?
- What is the clinician perspective on objective respiratory outcome measures for use in children under the age of five years admitted to PICU with a respiratory illness?
- What is the pathway of care for a child discharged from PICU?

4.2 Methods

4.2.1 Design

The study employed different qualitative methods in two phases: 1) semi-structured interviews using a survey approach, and 2) a focus group. The first phase employed semi-structured interviews with clinicians to describe the factors that clinicians identified as important when assessing HRQoL outcomes in children. Semi-structured interviews were considered an appropriate method in this exploratory approach to provide rich and illuminating data on clinician perceptions of HRQoL (Chapter 3).

The second phase employed a focus group methodology. Focus groups were chosen because they provide rich sources of insights and interpretations from participants (Polgar & Thomas, 1998). The group processes can also help participants to explore and clarify their views in ways that would be less easily accessible in a one-to-one interview (Kitzinger, 1996; Lane 2001). Group discussion was deemed particularly appropriate as the researcher had a series of open-ended questions on HRQoL which hoped to encourage participating clinicians to explore the issues of importance to them, in their own vocabulary, generating their own questions and pursuing their own priorities (Kitzinger, 1996). Disadvantages of focus groups include compromising the confidentiality of the research session, and the articulation of group norms may silence individual voices of

dissent (Kitzinger, 1996). Group dynamics or power hierarchies can affect who speaks and what a person says; it is also difficult or impossible to follow up the views of individuals (Robson, 1995).

4.2.2 Sample selection

A sample size of 6-20 participants is adequate for a qualitative study to develop clinician-generated items. A purposive sample of thirteen health care professionals was chosen in Phase 1 to represent clinical experts in general paediatrics and paediatric respiratory medicine/nursing, from tertiary, non-tertiary and primary care settings within the region. The principle of selection in purposive sampling is the researcher's judgement as to typicality or interest, and enables the researcher to satisfy their specific needs in a project. The rationale for the approach is very different from statistical generalisation from sample to population (Robson, 1995).

A purposive sample of three clinicians, representing clinical experts with specialist knowledge in paediatric respiratory medicine and physiotherapy, and one outcomes expert, representing an expert with specialist knowledge in developing outcome measures, were chosen for the focus group. Four participants were selected to represent those participants previously selected in Phase 1 to further explore and describe the concepts identified earlier. Focus groups generally comprise 4-10 participants (Kitzinger, 1996).

4.2.3 Materials

In Phase 1, a semi-structured interview schedule was used for the clinician survey to identify objective outcome measures in young children with respiratory diseases, and to identify the pathway of care of children discharged from PICU. The purpose of the latter objective was to identify possible mechanisms for accessing children to conduct HRQoL assessments.

In Phase 2, an open-ended question was used for the focus group interview: *"How do you determine the health status and HRQoL of a child (<5 yrs of age) with a respiratory illness?"* Other questions included, *"What subjective measures do you use?" and "What objective measures do you use?"*

4.2.4 Procedure

In Phase 1, all clinicians were contacted by letter or telephone and invited to participate in an interview. The interviews were arranged according to the clinicians' requirements, as a face-to-face interview, telephone interview or electronic-mail interview. Telephone interviews have the advantage of covering a large geographical area and generally obtain good response rates. They also allow for areas of uncertainty and ambiguity to be clarified, thus avoiding misinterpretations; and are less costly than face-to-face interviews (Cormack, 1991). However, face-to-face interviews have the advantage of allowing the interviewer to observe non-verbal cues. Using the Internet as a data-collection tool is less costly than face-to-face interviews and is fast; the data collected does not need transcription, which saves additional costs (Lakeman, 1997; Cooper, 2000). Electronic-mail is informal with a sense of equality (Eley, 1997). It offers the opportunity to clarify points and build a relationship with a respondent in a similar way to a telephone conversation, but this depends on the rapport that is established and the skill of the interviewer (Murray, 1995). Non-verbal cues that enhance the communication of emotion are more difficult to convey using electronic text only, but symbols can be used to convey emotions (Lakeman, 1997). Respondents to electronic-mail interviews can respond to questions at their convenience, but low response rates are common (Lakeman, 1997; Sell, 1997). People who use the Internet may also have certain characteristics from those who do not; a combination of data-collection methods in this study minimised the potential for demographic bias (Lakeman, 1997).

In Phase 2, the focus group interview was held in the research site, with topics for discussion introduced by the moderator who facilitated the contribution of the group participants (Polgar & Thomas, 1998). The meeting was audiotaped with the consent of the participants to facilitate data analysis.

4.2.5 Data analysis

Thorough notes were recorded of the main points raised during and after the telephone or face-to-face interviews in Phase 1 (Britten, 1996; Polgar & Thomas, 1998). The interview notes were coded using thematic analysis; during the coding process, concepts and themes and ideas were noted to form major categories (Polgar & Thomas, 1998). The audiotape from the focus group interview in Phase 2 was transcribed verbatim and also analysed using thematic analysis. The coding framework was theoretically informed by evidence on

HRQoL measurement, including dimensions of health described in existing HRQoL measures, in particular functional health.

4.3 Results

The sample composition in Phases 1and 2 are summarised (Tables 4.1 and 4.2).

Type of health care professional	Place of work
Consultant paediatrician (n=6)	Regional teaching hospital (n=2) District General Hospital (n=4)
General Practitioner – GP (n=4)	North Yorkshire (n=2) West Yorkshire (n=1) South Yorkshire (n=1)
Outreach Nurse (n=1)	Regional teaching hospital
Paediatric respiratory physiotherapists (n=2)	Regional teaching hospital

Table 4.1 Sample composition of clinician survey (Phase 1)

Membership of focus group	Institution
Consultant paediatrician (general paediatrics)	Research site
Consultant paediatrician (paediatric respiratory medicine)	Research site
Senior paediatric physiotherapist (paediatric respiratory / PICU)	Research site
Professor in Health Sciences (outcomes measurement expert)	Local university

Table 4.2 Sample composition of focus group interview (Phase 2)

4.3.1 Phase 1 - clinician survey

Interviews were conducted from 12th-30th April 1999. Six clinician interviews were conducted by telephone (four consultant paediatricians and two GPs); five clinicians were interviewed face-to-face (two consultant paediatricians, two paediatric physiotherapists, and one paediatric outreach nurse); and two interviews were conducted via electronic mail (both GP's). Thematic analysis of the clinician data revealed four main themes:

Theme 1: Assessment of respiratory outcome in children under the age of five years is reliant upon the subjective information gained from parents.

A major theme that emerged from the interview data was the subjective nature of the assessment of respiratory outcomes in this age group of children. All participants described how this assessment relied upon taking a clinical history from the child's parent. Thus

parental perception of the child's health and wellbeing was a major contributory factor to enable clinicians to assess the health and wellbeing of the child.

Two GP's asked parents about symptoms such as coughing and breathlessness, and the effects of these symptoms on the child's level of functioning, e.g. ability to feed, tolerate exercise, sleep at night. Sleep disturbance and coughing episodes were measures that two consultants used routinely. Both physiotherapists stated that they asked parents about the impact of the child's symptoms on the wellbeing and QoL of the child, e.g. the child's ability to keep up with their peers, and the effect of the child's symptoms on their daily activities. The physiotherapists assessed health status in more global terms than the medical staff, e.g. identifying impact of the child's illness upon family functioning and roles.

Theme 2: There are limited objective respiratory outcome measures used routinely in practice for children under the age of five years.

Difficulties in objectively assessing lung function in this age group were described by one consultant, who said that objective measures were available for children over the age of six years. There were differing opinions as to whether children under the age of five years could perform a peak flow test. Individual GPs said that this was possible in a three and four year old. Two consultants mentioned the measurement of lung function via age appropriate tests. The tests were not specified and were stated as being difficult to measure routinely in practice. Four consultants and both physiotherapists suggested that measures such as the number of medical consultations, number of GP prescriptions, and number of hospital admissions might give an indication of the child's respiratory status. However, they explained that even this was prone to bias, e.g. the reason for medical consultation may not be related to the respiratory illness, and owing to parental anxiety.

Two consultants, three GP's and both physiotherapists described how they took account of baseline values such as respiratory rate, pulse rate, oxygen dependency, and signs of respiratory depression (tracheal tug and intercostal recession) as measures of respiratory function. The outreach nurse described that oxygen saturation readings provided a measure of oxygen dependency. Medication use was described by one consultant as an objective measure; three consultants described how they asked parents about their child's use of

inhalers. The consultants were not currently aware of any objective respiratory outcome checklists or QoL measures that could be used in this age group.

Theme 3: Follow-up care is inconsistent for children discharged from PICU with a respiratory illness under the age of five years.

The follow-up care for this group of children was variable and inconsistent across the region. Several participants described that the health visitor (HV), GP, outreach nurse, or paediatrician in the local hospital outpatients department did not routinely see children. Other participants described more routine follow-up appointments with the GP, health visitor, outreach nurse and local paediatrician, for all children who were discharged from PICU. Some of the HV appointments were described as developmental checks, and therefore may not have been directly related to the child's reason for admission to PICU.

The physiotherapists described that if the child received treatment from a community physiotherapist prior to PICU admission, this child would still be seen by the community physiotherapist post PICU discharge. A small proportion of children (approximately 10%) are seen in the regional respiratory clinic at the regional hospital. Children with chronic respiratory problems were reported as seeing clinical nurse specialists, e.g. the asthma nurse, cystic fibrosis nurse, or neonatal outreach nurse. These visits took place at the child's home or in specialist local or regional clinics. The timing of the follow-up care was variable for those children that received it, two to four weeks, six weeks, or three months.

Theme 4: There is no optimal method suggested by healthcare professionals to obtain information on a child's health status and wellbeing.

Two consultants described that information on the child's health status and wellbeing may be gleaned from the child's hospital records, but often these are not very detailed or legible. One consultant suggested placing a sheet in the child's parent-held record, with a checklist on it for the health professional to complete at the next home or hospital visit. Two consultants and two physiotherapists suggested parent-held records as a means of obtaining objective information on the child. One GP stated that it would be possible for a parent to objectively record symptoms such as the number of episodes of nocturnal coughing, and sleep disturbance in a diary. Both physiotherapists suggested that the more objective measures of respiratory outcome might be recorded by nursery teachers and schoolteachers, such as developmental assessments. All consultants stated that it would be

difficult to remember individual children retrospectively to complete an objective checklist on respiratory outcomes. They described prospective, face-to-face interviews as a more suitable method of obtaining this information; GP's preferred to complete a short postal questionnaire.

4.3.2 Phase 2 – focus group interview

The focus group interview lasted 1.5 hours. Analysis of the group interview data using thematic analysis revealed six main themes about the use of a history-taking approach to assess the health status and HRQoL of a child. Participants identified specific questions that they asked regarding respiratory status (e.g. symptoms and outcomes), and general questions which they asked regarding health and wellbeing, such as physical, emotional, psychological, daily activities (hygiene, activity, sleep, eating and drinking, breathing, circulation, elimination, pain, anxiety, temperature, mobility, and communication), and social (family life, school, and play) questions. The five themes will be described in turn.

Theme 1: Identification of symptoms and clinical examination

Identification of symptoms

The participants described the identification of the presence of physical signs and symptoms of respiratory illness as important. For example, noisy breathing or a "ruttle", a local term (a wet noise due to secretions from the back of the throat), a wheeze (a whistling noise coming from the chest on breathing out), or a stridor (a rougher noise than a wheeze made on breathing in). These symptoms are assessed subjectively through discussions with parents. Both clinicians and physiotherapists agreed that the findings from auscultation of a child's chest were subjective. They also agreed that the assessment of the child's baseline was subjective and prone to recall bias, but GP records could be helpful. Information on how the child is currently compared to pre-admission is useful but difficult to assess objectively. The assessment of respiratory symptoms aims to identify if the child's lung function has improved or deteriorated. One of the clinicians explained that most of the symptoms that have been described are relatively common in young children anyway. So that 30% of all children under the age of twelve months will have recurrent coughing and wheezing.

Clinical examination

Clinical examination included assessing the child for signs of breathlessness (rapid breathing rate, shallow breathing, slow breathing rate, grunting, gasping, sternal or intercostal recession, nasal flaring, or cyanosis). If the child was breathless at rest then the child may have a significant respiratory problem. The clinicians ask parents if their child is breathless and if they are so breathless when exercising that they have to stop because they cannot keep up with other children the same age. The physiotherapist described breathlessness on feeding as being a particularly important sign of respiratory distress in babies less than twelve months of age. Snoring was also described as a symptom that is assessed.

One clinician explained that he asked parents about whether their child experienced pain; the other clinician said that he had never known a child under the age of five years to complain of pain on breathing. Clinical examination can also identify chronic respiratory problems such as a chest wall deformity - pigeon chest or Harrison sulci (permanent dents between the ribs). The child's gain in weight or growth (height) was described as possible objective measures that are determined on clinical examination. However, these measures are influenced by a number of factors; children who have chronic respiratory problems are likely to fail to thrive, but there may be other reasons for this such as behavioural feeding problems.

Theme 2: Effect of symptoms on the child's daily activities and family life

Daily activities

The functional significance of respiratory symptoms such as a wheeze was described as being much more important than the presence of the symptom, e.g. does the symptom affect activities during the day or waking up at night? The clinicians described difficulties in assessing exercise tolerance in young children and suggested that assessment of activity may be more appropriate. The physiotherapist described an assessment of the child's educational status, to determine if the child was able to go back to previous education, or if they needed a special school, and if their school attendance was better or worse. The ability of the child to take part in everyday activities without symptoms of breathlessness is also assessed.

Affect on daily activities

The physiotherapist in particular identified the importance of determining the affect of the child's symptoms on family life in terms of employment status, effect on other siblings, and financial effects. Also, if the parents' management of further episodes of illness has changed. The physiotherapist assesses if the parental relationships with the child have changed, e.g. is the child 'clingier' or are parents more distant from the child.

Theme 3: Drug treatments

Number

Clinicians identified the number of antibiotic prescriptions as a potential objective outcome measure. A number of respiratory children will be taking inhaled treatments, and some of them will be using their inhaler as and when they need it. One clinician stated that asking how many times a day children use their ventolin, bricanyl or atrovent inhalers (in this age group) will be important. If the child were taking their inhaler 10 times per day, this would suggest that their respiratory health status was poor.

Changes / adherence

The physiotherapist assesses the management of the child's condition at home since admission to identify any changes, e.g. more physiotherapy, better drug compliance, or better precautions to prevent further episodes.

Theme 4: Health care use

The clinicians described a possible index of severity to include number of hospital admissions post PICU discharge, number of hospital admissions for respiratory infections, and number of visits to the GP. The number of hospital admissions for respiratory infections were described as a more objective measurement of health status, but this is affected by wide variations in practice.

Theme 5: Family demographics and risk factors

Clinicians described the effect of environmental factors in this age group (cigarette smoking, damp housing, overcrowding, sibling number, and pets) that can exacerbate the respiratory symptoms. These factors may be considered as risk factors for respiratory disease.

Theme 6: Objective respiratory outcome measures

Clinicians identified potential objective outcome measures but they questioned the usefulness of these measures. The recording of oxygen saturation readings was mentioned as an objective measurement, but the clinicians reported that most children would have normal oxygen saturation reading post PICU discharge, and that this information is of limited value. A small minority of children will have low readings but these children will be receiving supplemental oxygen continuously. Clinicians explained that it would be difficult to measure oxygen saturation readings using a standardised test, but suggested that transcutaneous oxygen monitoring may be more reliable and cheaper method. Other objective measures such as chest X-rays were reported to be too crude an outcome measure; some children who have many respiratory symptoms have a normal chest X-ray, and some children who seem really quite well have a poor chest X-ray.

4.4 Discussion

The purpose of the study was to identify clinician's beliefs about PIC measures of effectiveness, including objective and subjective measures on outcome. Themes fell into similar categories as those found in Chapters 2-3, including the type and severity of respiratory symptoms and the impact of these symptoms on the child's daily life and activities. The main differences described by the clinicians were the language they used to denote signs and symptoms, some of which were different to parental descriptions. Items must therefore be phrased in the HRQoL measure in a way that parents can understand and answer. Clinicians also focused mainly on the physical dimension of a child's health, which is consistent with the HRQoL measures described previously (Chapter 2). The impact of the child's respiratory symptoms on the child's level of functioning in daily activities was also a prominent feature, reflecting emotional and cognitive dimensions of health. Emotional dimensions of health are also described in other child HRQoL measures, e.g. the HUI, AUQEI, PedsQL, QUALIN, Ontario CHSI and CAQ (Chapter 2). Few measures, however, describe cognitive dimensions of health, e.g. RAND, QoL in Nordic children, and the ITQoL measures (Chapter 2). The presence, severity and frequency of respiratory symptoms were also identified as a particularly important aspect of HRQoL from the clinician perspective. Symptoms have also been described in other respiratory HRQoL measures such as the CAQ (French et al, 1994b).

Traditionally clinician assessments are rooted in a belief that ill health is an objective measurable state, where poor health is a function of abnormality (biomedical model of ill health). The findings from the focus group interview indicated that although clinicians believed this they actually measured health in a more holistic and subjective way, including aspects of emotional and social wellbeing and functioning. This is particularly true for the physiotherapy assessment. Evidence suggests that proxy respondents less adequately measure the more subjective dimensions of health status than the more objective domains, which include functional capacity (Glaser et al, 1997a). Parents in particular tend to over-estimate a child's disability (Magaziner et al, 1988) and health care providers tend to underestimate a child's QoL (Sprangers & Aarson, 1992).

The pattern of follow-up care for children discharged from PICU was inconsistently reported. Individual clinician preferences and local resources may influence follow-up care e.g. facilities, personnel, and expertise. However, all children were seen by a healthcare professional post-PICU discharge, whether it was the GP, HV, outreach nurse or hospital doctor. No single optimal method for obtaining information on the respiratory health status and wellbeing of a child was described by the healthcare professionals. Methods of administration of an HRQoL measure, however, need to be responsive to the needs of the clinician in order to provide optimal uptake by them in routine clinical practice. While outcome measures, such as HRQoL measures, have been widely used in research to assess the effectiveness of interventions, they are rarely incorporated into routine practice (Greenhalgh et al, 1998). The barriers to the use of these measures within routine practice are complex, but at least a part of the problem is that clinical practice imposes specific demands on measurement that are not evident in research (Greenhalgh et al, 1998). The practicalities of using outcome measures in routine clinical practice must not be under estimated; practicality is considered one of the essential criteria of an outcome measure (Streiner & Norman, 1995).

The qualitative approach utilised was deemed appropriate for exploring in-depth clinician perceptions of HRQoL and respiratory outcomes in children under the age of five years with a respiratory illness. Determining the clinician perspective was essential in the development of an HRQoL measure to ensure its acceptability to clinicians and use in routine clinical practice to monitor and evaluate HRQoL outcomes. However, differing data collection methods were used in the clinician survey. Data obtained from the

electronic mail responses may not have been as rich and illuminating as data obtained from the telephone or personal interviews. A major problem with unstructured data collection techniques is that observer bias may cloud or distort the data being collected (Polgar & Thomas, 1998). The reliability of the data analysis may therefore be questioned in light of the differing data collection methods used. The participants interviewed were also not equally representative of the health professional groups they represented; there were more doctors interviewed than physiotherapists and nurses. It was therefore not possible to compare data between the different professional groups. The samples intentionally did not include representation from PICU clinicians, as clinicians from this specialty were represented on the RAG.

4.4.1 Developing clinician-generated items

The assessment of respiratory function and outcomes in this group of children is subjective in nature. Clinicians described symptoms such as breathlessness, tightness of the chest, and cough as measures of respiratory function. However, they recognised that these symptoms were all subjective health measures, and may be interpreted differently for the same child, by individual health professionals. Furthermore, the frequency and severity of symptoms is generally reliant upon reports from the child's parent or carer. Few objective respiratory outcome measures exist in everyday clinical practice. Those measures that were described, appear to be used inconsistently, and were dependent upon individual clinician preference, for example, peak flow readings. Few objective measures were identified from the literature; measures such as the interrupter technique are not used routinely in clinical practice. These objective measures may also be considered to provide a measure of pathophysiological state, rather than outcome per se. Measures such as peak flow readings are also variable and do not measure the underlying problem in a child.

The two qualitative studies carried out sets of interviews with clinicians to develop items for the HRQoL measure. Clinician-generated items are those based upon the questions that a clinician would ask if present and complement those items generated by parents (Chapter 3). Initially a clinician-generated index (CGI) was developed in five sections using data from the clinician survey, group interview, and a review of validated items from published generic and respiratory-specific HRQoL measures identified in the systematic review of child HRQoL measures (Chapter 2). The sections comprised items on the frequency and severity of respiratory symptoms; the effect of symptoms on daily activity; drug history;

health care use and family and home circumstances (Appendix VI). Clinicians identified the importance of knowing the medication that the child was taking and adherence to the medication plan. Items in the draft CGI that addressed this aspect however measure both patient outcome and medical process. Therefore, while a change in medication may well indicate deterioration in the child's health, it may represent no more than a change in the doctor's preferences. Similar difficulties may also arise with the CGI items on health care use; the danger with this section is that it measures both patient outcome and parental anxiety. While an increase in consultation rate may well indicate deterioration in the child's health, it may represent no more than a change in the parent's need for reassurance.

Most items in the draft CGI used binary responses and some items were based on a 3-point Likert scale. However, the item-scaling responses were later reviewed to include items with mainly five-point Likert responses (Chapter 5). The Likert-type response format has the advantage of allowing a middle 'undecided' response, but has the disadvantage that some respondents will give the middle response all the time (Polgar & Thomas, 1998). Most CGI items were based on a closed format for ease of data analysis and to minimise the time taken to complete the items (Robson, 1995). However disadvantages of this format include less 'depth' in answers, which may frustrate some respondents (Polgar & Thomas, 1998). A few items used an open-ended format to encourage more detailed answers, but with the added disadvantage of producing a less structured measure with responses that may be difficult to analyse and an increase in the time taken for the respondent to complete the measure (Polgar & Thomas, 1998).

4.5 Summary

Findings from both phases of the qualitative study identified that the routine assessment of respiratory function and outcomes in children under the age of five years is subject to clinician assessment using the methods of clinical history taking and clinical examination. This assessment is complemented by the information given to the clinician from the child's main carer or parent. Few objective respiratory outcome measures are routinely used, often in an inconsistent manner. Those that are used measure symptoms not health per se. Objective respiratory outcome measures described in the literature warrant further investigation.

Parental perception of the child's illness and symptom severity provides a subjective parent-based evaluation of the child's health assessment (Jenkinson, 1994). This parent-based evaluation forms the mainstay of the assessment of the child's health status and wellbeing, supplementing the clinician assessment, both of which are subjective. Clinicians focused mainly on the physical dimensions of health, but emotional and cognitive aspects were also considered. A CGI was developed reflecting these dimensions of health, incorporating medication and health care use, and home and family circumstances (risk factors). Subjective health measures can provide a valuable contribution to the assessment of illness in this patient group. They aid clinicians to develop treatment plans and to monitor the quality of medical care outcomes (Geigle & Jones, 1990).

CHAPTER 5 – Quantitative study to develop the HRQoL measure

5.0 Introduction

This chapter describes the development of the HRQoL measure, known as the PICQoL (Paediatric Intensive Care Quality of Life) questionnaire, from the item pool identified in Chapters 3 and 4 and theory reviewed in Chapter 2. A quantitative study (Phase II) to test the items for importance, agreement and dimensionality is described utilising a prospective and retrospective survey of parents whose children were discharged from a PICU (Streiner & Norman, 1995). Tests of homogeneity, internal reliability/consistency and dimensionality will be presented. The effect of the results in relation to the design of the PICQoL questionnaire will be discussed with implications for future validation.

5.1 Background

The development of measures requires the generation of items from a number of sources (Kessler & Mroczek, 1996). For example, patients or respondents themselves, clinical observation, theory, research, and expert opinion (Streiner & Norman, 1995; Juniper, Guyatt & Jaeschke, 1996). Once items have been generated from various sources, the scale developer is ideally left with far more items than will ultimately end up in the scale (Streiner & Norman, 1995). Various statistical techniques can be applied to select the best items from this pool and to determine the dimensionality of the HRQoL measure. These statistical techniques include the calculation of endorsement frequencies and item-total correlations (ITCs), and factor analyses (FA) (Streiner & Norman, 1995; Juniper et al, 1996).

In summary, the main areas identified from the parent and clinician perspective in relation to HRQoL are physical, social, emotional, behavioural, psychological and cognitive functioning, and respiratory-specific consequences such as the frequency and severity of respiratory symptoms and the impact of these symptoms on the child's daily activities. Similar dimensions of HRQoL were previously identified in the literature (Chapter 2).

5.1.1 Aim

This study aims to develop the PICQoL questionnaire using statistical techniques.

5.1.2 Objectives

- To identify those items that are redundant by computing endorsement rates, item-total correlations and factor analyses
- To test the internal consistency, homogeneity and dimensionality of the measure.

5.2 Methods

5.2.1 Design

A cross-sectional survey design was employed to test the PICQoL questionnaire. Questionnaires were employed in this development phase, both retrospectively and prospectively, and administered to parents, face-to-face. A cross-sectional approach is a suitable method for collecting descriptive information on HRQoL outcomes at one point in time (Creswell, 1994). A survey design provides a quantitative description of some fraction of the population, the sample, through the data collection process of asking questions of people (Fowler, 1993).

A survey design was chosen to provide a relatively simple and straightforward approach to the study of attitudes, values, beliefs and motives; to collect generalisable information from the population under study; and because of high amounts of data standardisation (Robson, 1995). This method also provides a rapid turnaround in data collection and is economical (Creswell, 1994). Data may, however, be affected by parental characteristics (e.g. their memory, knowledge, experience, motivation and personality); and parents may not necessarily report their beliefs and attitudes accurately (e.g. there is likely to be a social desirability response bias where people respond in a way that shows them in a good light) (Robson, 1995). Generally, no one mode of administration of a survey consistently outperforms all others (McColl et al, 2001). However, if multiple modes are necessary in order to ensure a high rate of participation in a project, and if change over time is a primary measure of interest, it might be best to keep the same data collection mode for any given respondent (Fowler, 1996).

Face to face administration of a questionnaire was chosen to allow the clarification of questions, and to encourage parental participation and involvement, and to increase response rates (Robson, 1995; Gotay, 1996). Non-verbal responses to a question could also be observed (Robson, 1995). This approach was utilised to evaluate parental perceptions of the questionnaire structure and layout, question wording and answer categories, an

important aspect of pilot work (Oppenheim, 1992). This approach also enables the identification of difficulties expressed by parents in understanding the items, whether due to a poor grasp of the language, limited intelligence, problems in concentration, or boredom (Streiner & Norman, 1995).

Face to face administration of a questionnaire may include the possibility of data being affected by the characteristics of the interviewer, such as experience as a health care professional, motivation, personality, skills, and experience (Robson, 1995). There may be interviewer bias, where attributes of the interviewer may affect the responses given, probably unwittingly, such as through verbal or non-verbal cues indicating 'correct' answer. Bias may also be apparent because of the interviewers' social or ethnic characteristics (Robson, 1995; Streiner & Norman, 1995). Respondents may also feel their answers are not anonymous and be less forthcoming and open (Robson, 1995). Bias was minimised by asking questions in the same way and handling unusual circumstances similarly (Streiner & Norman, 1995). Face to face administration may be costly, particularly if items need to be translated into one or more foreign languages and bi-lingual administrators found (Streiner & Norman, 1995); this was not a problem in this study as a bi-lingual interviewer was not utilised for financial reasons.

Retrospective studies require information about the past, which can be obtained from the participant's memory or from records. In the former case there is a real danger of bias, for memory distortions (recall bias) are a well-known phenomenon (Moser & Kalton, 1996). Therefore, a prospective approach, which has much to commend it over a retrospective approach, was utilised following the participants forward in time to minimise recall bias (Moser & Kalton, 1996). A prospective approach aimed to identify what was important to parents at the time of their child's PICU admission, rather than in retrospect. A retrospective approach does however have the advantage of speed with the results being available as soon as the data is collected (Moser & Kalton, 1996).

5.2.2 Sample selection

The aim of the sampling method is to draw a representative sample from the population so that one can confidently generalise from a representative sample to the rest of the population without having to take the trouble of measuring the rest of the population (Polgar & Thomas, 1998). If the sample is biased or not representative, it is difficult to

generalise validly from the sample to the population (Polgar & Thomas, 1998). The population included all UK PICU admissions satisfying the conditions over time of the study, namely children admitted to PICU with a respiratory illness under the age of five years. The sample selected in this study aimed to be representative of this population. Parents of children were selected according to the following inclusion criteria:

Retrospective sample

- Child was alive at time of sampling
- Child had a primary PICU admission diagnosis of a respiratory illness
- Child was <5 years of age
- Child had a PICU length of stay of ≥ 3 days
- Child was discharged from PICU between January 1999 and November 2000 (more than two months previously)
- Child had parents who were English literate

Prospective sample

As above, with the exception of the child's length of stay criteria, and two additions:

- Child had a PICU length of stay of ≥ 2 days
- Child's condition was stable, and not critical, at time of sampling
- Child was admitted to PICU between August and December 2000
- Child was still in hospital at time of sampling
- Parents were interviewed between Days 2-5 (48-120 hours) of PICU admission

Some children might have left the PICU by day five and so the sample may not be described as 'fully' prospective.

Parents of children from the larger PICU were selected as the number of children eligible to be sampled could be recruited from this site alone to meet the required sample size. Parents of children were selected retrospectively using the PICU computer database as the sampling frame (Chapter 3). The total population was used to represent the population under study, i.e. children admitted to PICU with a primary admission diagnosis of a respiratory illness, aged less than five years. A total prospective sample of parents whose child was admitted to PICU was also used.

5.2.3 Sample size calculation

Evidence in the literature on scale development for child HRQoL measures suggests that between 50-80 participants are thought to be adequate to test HRQoL items for reduction (Stein & Jessop, 1982; Cadman et al, 1986; Manificat et al, 1995; Collier & MacKinley, 1997; Ravens-Sieberer & Bullinger, 1998; Vogels et al, 1998). The sample size was also determined by other factors such as what was feasible for a single interviewer in terms of time and money (Oppenheim, 1992).

5.2.4 Questionnaire design

Questionnaire design is important as it affects response rate. Time spent on the planning of a questionnaire is pivotal to the final quality, not only of the measure, but also of the data obtained (Oppenheim, 1992; Bowling, 1997b). The PICQoL questionnaire was designed to evaluate HRQoL outcomes in children aged less than five years of age.

5.2.4.1 Question construction and response type for the HRQoL measure

Several factors were considered when designing the PICQoL questionnaire, including clarity, ambiguity, response options, and appearance. When developing items it is important to develop clear questions that are unlikely to be misconstrued by respondents (Kessler & Mroczek, 1996). In addition to inter-individual variation in the interpretation of the questions in QoL scales, it often happens that respondents differ in their understanding of response options (Kessler & Mroczek, 1996). Clarity of the items was ensured by asking questions appropriate to the reading level skills of a 12-year old (Streiner & Norman, 1995). The readability scores of the prospective and retrospective PICQoL questionnaires were tested using the facility in the word-processing software (Microsoft Word). Results revealed acceptable reading ease and reading grade statistics (68.9% and 6.0 for the retrospective questionnaire; 59.8% and 7.9 for the prospective questionnaire).

Ambiguity in the question wording and response alternative was avoided by ensuring that double-barrelled questions, where two questions are asked at the same time can be answered differently, were not used (Streiner & Norman, 1995; McColl et al, 1998). The use of jargon, idioms or metaphors was also averted, which can also aid future translation if required (Streiner & Norman, 1995; Juniper et al, 1996). Value-laden and positive or negative wording was also avoided; negatively worded items tend to have lower validity coefficients than positively worded ones (Streiner & Norman, 1995). Double negatives

were also avoided, e.g. a negative statement followed by a disagree response (McColl et al, 2001). Questions were kept short, aiming ideally for a sentence of less than twenty words (McColl et al, 2001). Consideration was given to the ordering of questions in the questionnaire to achieve a smooth, logical flow of ideas (Jack & Clarke, 1998). The questionnaire started with easy, non-threatening questions; the general questions preceded the specific questions (Robson, 1995; Jack & Clarke, 1998; McColl et al, 2001). Writing clear items is a sorely underrated challenge; it is extremely difficult to phrase questions that are clear and will be understood in a similar manner by all respondents (McDowell & Jenkinson, 1996).

Questions need to be asked of respondents over a well-defined period of time, and a period of two weeks is commonly used to provide an accurate recall (Juniper et al, 1996). However, others recommend that questions can be reliably asked in relation to the preceding six months, unless they are asking about specific events, as over time most people's memory becomes blurred (Bowling, 1997a). Many questions in the PICQoL questionnaire were worded to consider a child's health and wellbeing in the previous two weeks, other questions asked parents a present assessment, e.g. feelings of anxiety at this moment.

A Likert-scale response option was selected over a visual-analogue scale, as it is easier to construct, administer and interpret than other scales (Oppenheim, 1992; Juniper et al, 1996). The reliability of Likert scales also tends to be good, partly because of the range of answers permitted to respondents (Oppenheim, 1992). Likert scales also provide more precise information about a respondent's degree of agreement or disagreement, and respondents usually prefer this to a simple agree/disagree response (Oppenheim, 1992). It also becomes possible to include items whose manifest content is not obviously related to the attitude in question, enabling subtler and deeper ramifications of an attitude to be explored (Oppenheim, 1992). However, Likert scales have been criticised for their lack of reproducibility in the technical sense; the same total score may be obtained in many different ways (Oppenheim, 1992). The pattern of responses therefore becomes more interesting than the total score (Oppenheim, 1992); this is particularly so for clinicians interested in evaluating HRQoL outcomes in children discharged from PICU. Another criticism has been that since the scale offers no metric or interval measures, it is not

possible to know where scores in the middle ranges change from mildly positive to mildly negative (Oppenheim, 1992).

Often investigators choose a seven-point scale when developing an evaluative HRQoL measure, which must be responsive to important changes even if they are small (Juniper et al, 1996). Up to a point, and no one is yet sure of what that point is, increasing response options on a scale will increase item responsiveness (Kirshner & Guyatt, 1985). That is, individual items will show changes in score when clinically important improvement or deterioration occurs (Kirshner & Guyatt, 1985). McColl et al (2001) report that it has been suggested that increased precision may be achieved through the use of seven rather than five response categories, especially in Likert-type scales; however, they suggest that there is little evidence for further enhancement of precision beyond seven categories. Further research into the reliability and discriminatory power of five versus seven point (or more finely graded) scales is recommended (McColl et al, 2001).

Likert-scales were used in the development of clinician-generated items (Chapter 4), and parent-generated items. The most common response option, with five-points, was favoured in the development of items in the PICQoL questionnaire, as they allow for a neutral category or midpoint (Oppenheim, 1992; Robson, 1995). Robson (1995) reports that there is disagreement on the wisdom of including a 'middle alternative'. On the one hand it may encourage a non-committal response, on the other it allows for an additional gradation of opinion. Typically, 20% of respondents may use the middle category, but it appears that its inclusion or exclusion does not affect the relative proportions of those actually expressing opinions (Robson, 1995). McColl et al (2001) report that the middle response category in attitude/opinion questions does not necessarily represent a position of neutrality, so it should be included. The subjective rating scales used in the PICQoL questionnaire were placed on separate pages to offset the 'halo effect' (Oppenheim, 1992).

Attention to the appearance of the questionnaire, including its length and layout, was considered as this may reduce the perceived or actual burden of response (McColl et al, 2001). Through good design, the risk of errors in posing questions and coding responses can be reduced and potential variability between interviewers or coders minimised, thus reducing bias (McColl et al, 2001). Appearance of the questionnaire can influence the respondent at several stages, including the arousal of interest to complete the questionnaire,

evaluation of the task involving perceptions of time and effort required to complete the questionnaire, and finally initiation and monitoring the task of completion which identifies the actual burden of response (McColl et al, 2001). Colour, brightness, and different font styles were used in a consistent manner to navigate respondents through the questionnaire.

5.2.5 Item generation

The main themes elicited from parental data included the impact of the child's respiratory symptoms on the child's growth and development, daily activities, and family life (Chapter 3). Items were developed to reflect the growth and developmental milestones of children under the age of five years. It was deemed inappropriate to develop age-appropriate items, as this would have led to several versions of the questionnaire needing to be developed, which was not feasible. Items were developed to represent the main developmental milestones within the specified age-range; these items were informed by the literature on child development and the author's clinical experience (Davenport, 1994). Items were worded such that parents compared their child to other children the same age as their child. Using the words *'in comparison with other healthy children the same age as your child'* as the frame of reference for respondents aimed to minimise parental misunderstanding, avoid ratings diverging more than they might have done, and facilitate interpretation (Oppenheim, 1992; Jack & Clarke, 1998). If the child was too young to achieve the stated milestone, the parent could respond with a 'not applicable' response option. Clinicians in the RAG verified the face validity of these items.

Parents described positive and negative consequences of the PICU illness for their children; items were developed to measure these consequences. Consequences included changes in a child's developmental milestones, particularly in relation to growth (height and weight), physical (motor), emotional (positive and negative), social (interaction) and cognitive (understanding) functions. Parents also described pain and behavioural consequences. Items were developed to reflect these consequences from the parental perspective.

The impact of the child's health on family life was also an important finding from the parental data, and was particularly emphasised by those parents whose children had a chronic illness. These parents described the effect of their child's health on family life in terms of the inability to go on an outing without prior planning, or not being able to go out

at all, and general disruptions to family life. Parents described the impact of their child's health on their working life in terms of not being able to work or having to reduce their working hours because of caring for their sick child. Parents also described the impact of their child's health on their relationship with their partner in the theme of 'negative consequences'. Parents particularly emphasised the inability to spend time with their partner or to go out socially. Partner relationships were described as a category under the theme of 'stressors' (factors causing parents stress during their child's PICU illness). A review of other generic child HRQoL measures, in particular the CHQ, also revealed a similar dimension of health on family activities and family cohesion (Landgraf et al, 1996). The items comprising this dimension of health in the CHQ were reviewed and were considered appropriate to include within the PICQoL questionnaire; permission was gained from the CHQ developer to include these items.

5.2.5.1 Measures for other aspects of the parental consultation

Two themes that emerged from the parental interviews were illness perceptions and worry/anxiety. There are standardised measures to evaluate these two concepts. There is currently only one measure to evaluate illness perceptions, the Carer-version of the Illness Perception Questionnaire (IPQ); this was used (Weinman et al, 1996). The IPQ is a theoretically driven questionnaire, based on a self-regulatory model of illness representations, which in turn determines coping (Leventhal, Nerenz & Steele, 1984). This model is described previously (Chapter 3). The IPQ items were derived from previously described symptom checklists, and interviews with patients; some were generated by the scale developers (Weinman et al, 1996).

The Carer-IPQ comprises five illness representation components: identity (12 core symptoms); cause (10 items); timeline (4 items); consequences (7 items) and cure/control (5 items). Timeline items measure a patient's perception of the length of their illness. In the identity scale the carer is asked to rate how much they feel the symptoms listed are part of their spouse/partner's illness on a four-point scale ranging from 'all the time' to 'never' according to how often each symptom is experienced as part of the patient's illness. The identity scale is scored by summing the number of items endorsed at 'occasionally' or greater, so that the total score ranges from 0-12 for the core list (Weinman et al, 1996). The timeline, consequences and cure/control scores are presented in a mixed order and are rated by the carer on a five-point scale ranging from 'strongly disagree' to 'strongly agree'

(scored 1-5). After reverse scoring appropriate items, scores for timeline, consequences and cure/control are obtained by summing all the scale items and dividing by the number of items (Weinman et al, 1996). It is not appropriate to sum all the items in the cause scale as each item represents a specific causal belief, but items may be combined to identify internal and external causal factors (Weinman et al, 1996).

The psychometric properties of the general IPQ scale are reported favourably, with good levels of internal consistency (Cronbach's alpha 0.73-0.82), and good test-retest reliability, tested at one, three and six months (Weinman et al, 1996). Concurrent, discriminant and predictive validity of the IPQ is also reported encouragingly (Weinman et al, 1996). The IPQ has been tested in several field trials in adult patients with chronic obstructive airways disease (Scharloo et al, 2000a & 2000b). The Carer-IPQ has been tested in only one study of 50 adult myocardial infarction patients and their spouses (Weinman et al, 1996). Significant intercorrelations were found for consequences, cure/control and timeline scores, but not for the identity scale. Evidence on the reliability and validity of the Carer-IPQ is limited to this study. However, in view of good evidence of the psychometric properties of the general IPQ, the Carer-IPQ was chosen for inclusion in the HRQoL measure.

Weinman et al (1996) state that the core list of items in the identity scale may be added to by researchers to tailor the scale to specific illnesses; and each item in the IPQ may be adapted to replace 'illness' with the name of a particular illness, e.g. 'asthma' or 'diabetes'. The Carer-IPQ was adapted to include a symptom list relevant to childhood illnesses, and the item wording adapted to read *"My child's PICU illness"* instead of *"My partner's illness"*. These modifications were piloted with some further clarification required (see Section 5.3.1).

Items were developed within the PICQoL questionnaire to evaluate parental perceptions of worries about their child's physical health, emotional state, behaviour, and learning abilities. In addition two standardised measures were reviewed for evaluating parental anxiety: the short-form State Trait Anxiety Inventory (Spielberger, Gorsuch & Lushene, 1970; Spielberger, 1983), and the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983).

The short-form State Trait Anxiety Inventory (STAI: Y-6 item) was developed from one of two STAI questionnaires (Marteau & Bekker, 1992), Form X and Form Y (Spielberger et al, 1970; Spielberger, 1983). The original STAI was developed from an item pool of 177 questions taken from existing anxiety scales (Spielberger, 1983). Extensive testing of items was carried out largely on college students (n=5000+); the final version was subjected to item and factor analyses (Bowling, 1995). Spielberger's long-form scale comprises twenty items measuring current anxiety (State Anxiety) and the disposition to respond to stressful situations with high anxiety (Trait Anxiety) (Riddle et al, 1989). Each state item is rated on a four-point intensity scale, from 'not at all' to 'very much so' and each trait item is rated on a four-point frequency scale from 'almost never' to 'almost always' (Bowling, 1995). Two scores are calculated, state anxiety and trait anxiety, ranging from 20-80 with high scores reflecting greater levels of anxiety. The STAI (long-form) is reported to have good construct validity, internal consistency and test-retest reliability. The measure is self-administered and takes less than ten minutes to complete (Spielberger, 1983). There is a version of the STAI for children, which has the same 20:20 item format for trait and state as the adult version, and is reported to have good results for reliability and validity. The child version of the STAI was inappropriate for this study, as children were not completing the measure.

The long-form STAI (Spielberger et al, 1970) has been used to assess the overall anxiety response and the propensity for parental stress in a PICU setting (Eberly et al, 1985; Riddle et al, 1989). The mean scores for both mothers (52.76) and fathers (48.71) fell within the category of 'anxious clients' (Riddle et al, 1989). Carnevale (1988 & 1990) proposes that state anxiety should be utilised as a non-specific indicator of coping efficacy.

A short-form STAI has been developed and tested in various groups over the last ten years (Bekker et al, 2003), including four groups of participants (38 medical students, 45 student nurses, 200 pregnant women, and 23 pregnant women with abnormal routine screening tests) (Marteau & Bekker, 1992). The short-form is reported to have acceptable reliability (internal consistency) and produced scores that were similar to those produced with the full-form across subject groups manifesting normal and raised levels of anxiety (Marteau & Bekker, 1992). Concurrent validity was assessed against the full-form and there were no differences in the mean scores on both forms for all groups. Marteau & Bekker (1992)

report that test-retest reliability was not assessed as test-retest reliability is low for state anxiety because of state anxiety's transitory nature.

The HAD scale comprises fourteen items on two sub-scales, anxiety and depression (Zigmond & Snaith, 1983). Ratings by participants are made on four-point scales which represent the degree of distress: none = 0, a little = 1, a lot = 2, and unbearably = 3; items are summed on the two sub-scales (Zigmond & Snaith, 1983). High scores indicate the presence of problems (non-cases = 7 or less; doubtful cases = 8-10; definite cases = 11+) (Zigmond & Snaith, 1983). Initial tests of reliability and validity are reported as good, and scale scores were not affected by the presence of physical illness; however, test-retest reliability was not assessed (Zigmond & Snaith, 1983). The HAD scale is particularly useful for measuring the outcome of physical conditions owing to its omission of somatic items, but further testing is required of its reliability and validity before its performance as an indicator can be confidently judged (Bowling, 1995 & 1997a). It is also recommended that the HAD scale be interviewer-administered (Bowling, 1995).

The short-form STAI: Y-6 item was chosen for inclusion in the HRQoL measure over the HAD because of good evidence of its psychometric properties, including feasibility and clinical utility. The STAI is one of the most widely used measures of anxiety in psychological and clinical research (Bowling, 1995). It is also practical, comprising six items, which can be completed in less than a few minutes. The HAD is interviewer-administered which is not feasible within this research programme as the HRQoL measure is intended for self-administration; the HAD was rejected on this basis.

5.2.5.2 Demographic items

Items on child and parent demographics were developed based upon a review of other child HRQoL measures and clinician requirements. Demographic items utilised in the Office of Population Censuses and Surveys (OPCS) questionnaires were reviewed, and those from a 'Lifestyle Survey' covering the former Yorkshire Region (NHSE, 1995). Developed items included those on child and parental sex, child and parental age, family history, parental ethnic origin, parental marital status, parental educational status and parental employment status and change in employment. Items were also generated on the child's previous use of health care services, past medical history and risk factors for respiratory disease (Chapter 4). The child's gestational age was also considered when

parents completed the developmental milestone items, and an explanation was given regarding this at the start of the relevant items, following the advice of an expert clinician (consultant neonatologist).

5.2.5.3 Assessment of illness severity

Items were also generated from clinician data on the child's illness severity, including the frequency and severity of respiratory symptoms (Chapter 4).

5.3 Materials

The PICQoL questionnaire was developed with reference to empirical data described in Chapters 2-4, and to literature on the development of child HRQoL measures. The IPQ and short-form STAI were incorporated within the PICQoL questionnaire as described previously. A retrospective and prospective version was produced for pilot testing. The prospective version was slightly shorter than the retrospective version as some items were removed because they were inappropriate to ask prospectively, e.g. comparing pre and post PICU general health state and respiratory health state.

5.3.1 Pilot questionnaire

The design of the PICQoL questionnaire was an iterative process performed in consultation with clinicians and research colleagues from the RAG. The PICQoL questionnaire was piloted on a small group of retrospective (n=12) and prospective (n=2) parents via face-to-face administration. Piloting aimed to improve questionnaire clarity and remove any problems, before the main study (Chapter 6) (Polgar & Thomas, 1998). The pilot respondents were also asked whether the questions were clear (Polgar & Thomas, 1998).

Findings from field notes recorded during the pilot study revealed that some modifications were required to aid questionnaire clarity and improve comprehensiveness. The layout of questions on frequency and severity of respiratory symptoms were amended by integrating the frequency and severity items for each symptom, rather than presenting all frequency items separately then all severity items. An additional symptom of a 'runny nose or snuffles' was included in the respiratory symptoms section. The list of symptoms in the Carer-IPQ was revised to include more child-appropriate symptoms, such as 'cough', 'runny nose' or 'snuffles' and 'fever'. More adult oriented items, such as 'stiff joints' and 'headaches', were removed. Eleven items (IPQ symptom list and child's past medical

history) were added to the measure following piloting, and two items were removed from the IPQ symptom list, as they were not child-appropriate. Clarification was needed for some parents on the Carer-IPQ in relation to their child's PICU illness; some parents of children with a chronic illness found it difficult to separate perceptions of their child's PIC illness from those of their child's chronic illness.

Several parents identified that their children were born prematurely. Prematurity is considered a risk factor for respiratory disease, and so an item was added to assess the child's prematurity via their gestational age. This item might indicate chronic problems associated with prematurity. Parents found it difficult to assess their child's development if their child was born prematurely; they knew that compared to other children of their child's age, their child was likely to be behind in their development. Items were included on maternity history and a statement made about the child's corrected gestational age to assist parents in completing the daily activities items. Items on the child's daily activities were revised to encompass statements appropriate to babies, infants and toddlers; an item on the child's weight was added. An item on the presence of an acute or chronic illness was added to aid known groups comparison (discriminant validity).

Generally parents commented positively on the questionnaire design but boxes for the past medical history items were made clearer. Items on 'cancer', 'kidney problems', 'liver problems' and 'neurological problems' were added to the past medical history list on the recommendation of clinicians from the RAG. The 'employment status' item was removed in the parent demographic section and replaced with an item on 'changes to work status' as clinicians believed this was more relevant. An 'impact on family life' item, 'stopped you/your partner going to work' was removed, as some parents did not work. The retrospective and prospective questionnaires were modified in light of the pilot findings (Appendices VII and VIII).

5.4 Procedure

In March 1999, LREC approval was given for the retrospective, face-to-face administration of the PICQoL questionnaire and further LREC approval was given in August 2000 for the prospective, face-to-face administration of the PICQoL questionnaire. General practitioners of children sampled retrospectively were contacted to confirm that the child was alive and well before parents were contacted by letter and invited to

participate in the study, with or without their partner, at home or in hospital. Conducting the study in the parents' home aimed to put the parent at ease, since they are in familiar surroundings, and to increase compliance, because parents might not want to travel (Streiner & Norman, 1995). However, this method involves greater cost to the investigator and the possibility of interruptions, e.g. by telephones or family members (Streiner & Norman, 1995). The letter contained information about the study and a reply slip as described previously (Chapter 3). The author contacted parents by telephone to arrange the questionnaire administration; travelling expenses were reimbursed to parents who chose to participate at the hospital.

The author approached prospective parents on the PICU, after discussion with the family care nurse and/or child's nurse as to the appropriateness of approaching the parent for informed consent. An information sheet was given to retrospective and prospective parents, and written informed consent, including permission to obtain information from other health care resources, was obtained. The documentation used was similar to that described previously (Appendix III). The author returned to prospective parents within 24 hours to administer the questionnaire. Prospective questionnaire administration was conducted in an interview room or at the child's bedside or on the children's ward following the child's PICU discharge. Follow-up was offered to all parents by inviting them to obtain a summary of the research results by completing a relevant section on the consent form (Chapter 3). Prospective parents were interviewed 48-120 hours after their child's admission to the PICU. The author read out each item on the prepared PICQoL questionnaire and parent(s) indicated their response on their own copy.

The child's illness type was classified as either an acute respiratory illness (e.g. bronchiolitis), or an acute respiratory illness with underlying chronic respiratory condition (e.g. pneumonia with chronic lung disease/asthma), or an acute respiratory illness with a non-respiratory chronic condition (e.g. chest infection with cerebral palsy/muscular dystrophy). The classification of illness type was determined by the author independently from the admitting clinician using information from the sampling frame. Definitions of acute and chronic were verified with experts in the field of respiratory medicine.

5.5 Data analysis

The HRQoL measure is a structured questionnaire with tick boxes. Data was entered into a statistical computer package SPSS version 10.0 (SPSS Inc. 1999), as this was easily available to the researcher. The pilot sample was included in the main sample for data analysis; and missing pilot items for 'weight' (daily activities question) and 'runny nose or snuffles' (respiratory symptoms question) were coded using median values. Missing pilot demographic items were excluded from the main analysis. Generally, missing data was replaced by median values in the calculation of item-total correlations of daily activities items, respiratory symptom items, and IPQ items; and the FA (factor analysis) of daily activities items. However, in the FA of IPQ items, missing values were replaced with mean values. Itemwise replacement of missing values using population means or medians was utilised if less than 50% of the total number of items comprising a PICQoL score were missing per case.

A 10% sample of the entered data was cross-referenced against the original questionnaires (sample edit). Omission and consistency editing checked for missing data and the consistency of responses, so encouraging confidence in the final results. The randomly selected and total samples used aimed to be representative of the PICU population of children aged less than five years admitted with a respiratory illness. The data analytic methods employed are presented in four sections: demographics including standardised measures; selecting items for the scale - item reduction; homogeneity of the measure incorporating internal consistency; and dimensionality of the measure.

5.5.1 Demographics and standardised measures (STAI and IPQ)

Descriptive statistics were used to identify frequency distributions, statistics of central tendency and dispersion for each question (Fowler, 1993; Moser & Kalton, 1996). Statistical data analysis of IPQ items aimed to calculate symptom (illness identity), timeline, consequences and cure/control scores; STAI scores were also calculated. Sample differences were explored using t-tests of IPQ and STAI scores (Clegg, 1990).

5.5.2 Practicality

Practicality of the PICQoL questionnaire was assessed by an item within the questionnaire asking parents to rate how easy they found it to complete the questionnaire on a five-point Likert scale ranging from 'very easy' to 'difficult'. Parents were also asked to record

questionnaire completion times to assess practicality. Descriptive statistics summarised practicality of the PICQoL questionnaire.

5.5.3 Selecting items for the HRQoL measure (item reduction)

Endorsement rates, item-total correlations (ITCs) and factor analyses were the statistical techniques chosen to select items for inclusion in the PICQoL questionnaire (see Section 5.5.5 for a description of FA). If items met the inclusion criteria for endorsement or ITC they were included in the PICQoL questionnaire. Exceptions to this rule were those items that did not meet these criteria but for theoretical or clinical reasons warranted inclusion. Endorsement rates were calculated by computing the proportion (p) or percentage of people responding to each response alternative. Items were included within the PICQoL questionnaire when the endorsement rate of at least one response alternative was between 02-0.8, and none of the possible alternative responses had an endorsement rate greater than 0.8 (Streiner & Norman, 1995).

5.5.4 Internal consistency and homogeneity of the measure

In order for a scale to be homogenous, all the items should be tapping different aspects of the same attribute and not different parts of different traits (Streiner & Norman, 1995). Tests of homogeneity and internal consistency were conducted: items should be moderately correlated with each other, and each item should correlate with the total scale score (Streiner & Norman, 1995). Inter-item correlations were calculated using Cronbach's alpha (α) statistic (Cronbach, 1951). Cronbach's alpha statistic can be used on scales where there are more than two response alternatives (Jenkinson & McGee, 1998). If alpha values increase significantly when a specific item is left out, this indicates that the exclusion of the item increases the homogeneity of the scale. Alpha statistics of 0.7-0.9 are generally included; those below 0.5 suggest that items are not all tapping the same underlying area of interest (Nunnally, 1978; Streiner & Norman, 1995; Jenkinson & McGee, 1998). Items with lower correlations can be discarded (Kline, 1986) or rewritten (Streiner & Norman, 1995). The remaining items can be selected, starting with the highest correlation (Streiner & Norman, 1995). Alpha coefficients above 0.9 indicate that a measure may have high internal reliability and can be used at the level of individual analysis (rather than only at the level of group analysis) (Jenkinson & McGee, 1998). However, some would suggest that such high alpha values would indicate that effectively

the same question is being asked more than once (Jenkinson & McGee, 1998). Cronbach's alpha statistic gives an indication of the internal consistency or reliability of a measure.

Corrected item-total correlations were calculated using the Pearson product-moment correlation coefficient (or Spearman's rank correlation coefficient for non-normally distributed data). Corrected item-total correlations describe the correlation of the individual item with the scale total omitting that item (Streiner & Norman, 1995). Items with correlations <0.20 are generally discarded from a measure (Streiner & Norman, 1995).

5.5.5 Dimensionality of the HRQoL measure

Dimensionality of the measure was assessed using exploratory FA in which the relationships between various variables are examined without determining the extent to which the results fit a particular model (Bryman & Cramer, 2001). This analysis attempts to identify underlying variables, or factors, that explain the pattern of correlations within a set of observed variables (Kinnear & Gray, 1995). Factor analysis is a method for simplifying complex sets of data and is usually applied to correlation matrices (Kline, 1994). Ideally any test should measure only one variable. To ensure this a large number of possible items are administered to participants and the correlations between the items are subjected to FA. An item should 'load on', be correlated with, the scale it belongs to, and not any other one. If it loads on the 'wrong' factor, or on two or more factors, then it is likely that it may be tapping something other than what the developer intended, and should be either rewritten or discarded (Streiner & Norman, 1995). Items which load the general factor are selected for the test or measure; FA therefore aids item selection (Kline, 1994).

The two main types of FA are principal-components analysis (PCA) and principal-axis factoring (PAF). In PCA, all the variance of a score or variable is analysed, including its unique variance (combination of specific and error variance) (Bryman & Cramer, 2001). In PAF, only the variance that is common to or shared by the tests is analysed; an attempt is made to exclude unique variance from the analysis (Bryman and Cramer, 2001). The philosophy of the two approaches is different. Principal components analysis is more practically based and useful when applying a questionnaire in its present form to other respondents to obtain the best summary of the information obtained in practice. However, PAF is useful for exploring underlying theories and concepts, and this approach was used to assess the dimensionality of the HRQoL measure in the development phase. Varimax

(orthogonal) rotation (Kaiser, 1958) is recommended as this aims at simple structure with the advantage that the factor loadings are equivalent to the original analysis and the actual factors are being dealt with (Kline, 1994).

Ideally FA should be performed separately for retrospective and prospective questionnaires, including analysis of the IPQ items. However, owing to the small sample size this is inappropriate and so FA was performed on merged retrospective and prospective data. There is no consensus on what the sample size for FA should be, but it has been proposed that the minimum sample size is five participants per variable and no fewer than 100 individuals per analysis (Bryman & Cramer, 2001).

5.6 Results

5.6.1 Sample representativeness

Retrospective

Utilising the entry criteria of admission to PICU with a respiratory illness, a length of stay of more than or equal to three days and a child's age of less than 1825 days (5 years), 119 PICU admissions were identified from the sampling frame for the retrospective sample between 1st January 1999 and 15th July 2000. However, this number was reduced for several reasons (Table 5.1).

Reason for exclusion from sampling frame	Number of admissions
Child died during or following PICU discharge	30*
Duplicate PICU admissions (excluding deaths)	6
GP refused permission to contact parent	1
Child/parent moved away from region	1
Error in child's date of birth entry in sampling frame	1
Child was one of twins (therefore same parent)	1
Child was sampled in Phase I	12†
Total	**52**

*Represents 21 children.
†Represents 10 children

Table 5.1 Reasons for exclusion from the sampling frame (Phase II)

With the above exclusions, parents of 67 children (119-52) were eligible to be sampled following discharge from PICU 2-19 months previously (5th January 1999 – 4th November 2000). A total sample of parents representing 67 children was therefore sampled. Two

letters were returned by the Post Office as undeliverable, thus reducing the sample size to sixty-five. Responses were received from one parent representing 40 children (39 mothers and one father), a response rate of 61.5%; a further seven fathers also participated in the retrospective interviews. Questionnaires were administered to parents between June-October 2000. Non-responders to a written invitation to complete a questionnaire retrospectively were telephoned at home to promote the recruitment rate. Seven of the 40 mothers who participated did so in response to a follow-up telephone invitation.

Prospective

Twenty-five parents representing seventeen children were interviewed prospectively between 1st August 2000 and 2nd December 2000. The sample comprised seventeen mothers and eight fathers. During the timeframe, seven children were excluded from the prospective interviews (Table 5.2); the response rate was therefore 70.8% for one parent participating. The prospective interviews represented 34.7% of all interviews (n=72) conducted in this phase. Parents were interviewed retrospectively at home (n=42) or hospital (n=29); one parent preferred to complete a questionnaire by postal response.

Reason for exclusion	Number of children
Long-term patient (outside 2-5 days post PICU admission time period)	1
Parents did not want to participate	1
Child's length of stay < 2 days	2
Child's diagnosis not respiratory	1
Possible withdrawal of child's treatment	1
Parents non-English speaking	1
Total	7

Table 5.2 Reasons for exclusion from prospective interview (Phase II)

5.6.2 Parental demographics

Results will be presented separately for retrospective and prospective parents. Respondents included 47 retrospective parents and 25 prospective parents; most respondents were female and the child's mother (Tables 5.3-5.4).

Sex of respondent	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Female	39 (83)	17 (68)	56 (78)
Male	8 (17)	8 (32)	16 (22)

Table 5.3 Parental sex (Phase II)

Relationship of respondent	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Biological parent	44 (94)	24 (96)	68 (94)
Step-parent	1 (2)	0 (0)	1 (1)
Foster parent	2 (4)	1 (4)	3 (4)

Table 5.4 Relationship of respondent to child (Phase II)

Most parents were Caucasian, married and attained GCSE or O levels as their highest grade of educational qualification (Tables 5.5-5.7).

Ethnic origin of respondent	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Caucasian	38 (81)	22 (88)	60 (83)
Black	1 (2)	0 (0)	1 (1)
Indian	1 (2)	0 (0)	1 (1)
Pakistani	7 (15)	3 (12)	10 (14)

Table 5.5 Parental ethnic group (Phase II)

Marital status	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Married	37 (79)	18 (72)	55 (76)
Living together	6 (13)	5 (20)	11 (15)
Divorced or separated	1 (2)	0 (0)	1 (1)
Single	3 (6)	2 (8)	5 (7)

Table 5.6 Parental marital status (Phase II)

Highest educational qualification attained	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
No formal education	11 (23)	6 (24)	17 (24)
GCSE or O level	21 (45)	10 (40)	31 (43)
A Level	5 (11)	0 (0)	5 (7)
Professional qualification	3 (6)	1 (4)	4 (6)
Degree or higher	7 (15)	8 (32)	15 (21)

Table 5.7 Highest grade of parental educational qualification (Phase II)

The mean parental age from the retrospective sample was 30.8 years (SD 5.7 years; range 19-50 years) (Figure 5.1) and for the prospective sample was 31.0 years (SD 5.4 years; range 20-46 years) (Figure 5.2). The overall mean parental age for all respondents was 30.9 years (SD 5.56 years; range 19-50 years) (Figure 5.3). The distribution of parental age is near normal symmetry with the exception of one parent aged 50 years.

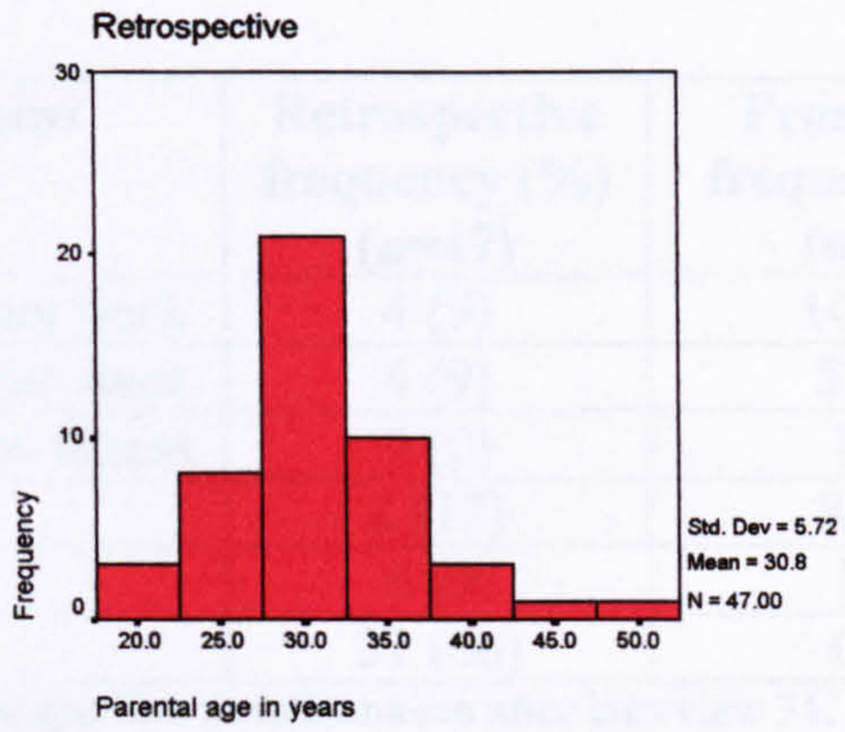


Figure 5.1 Histogram of parental age in years (retrospective sample) - Phase II

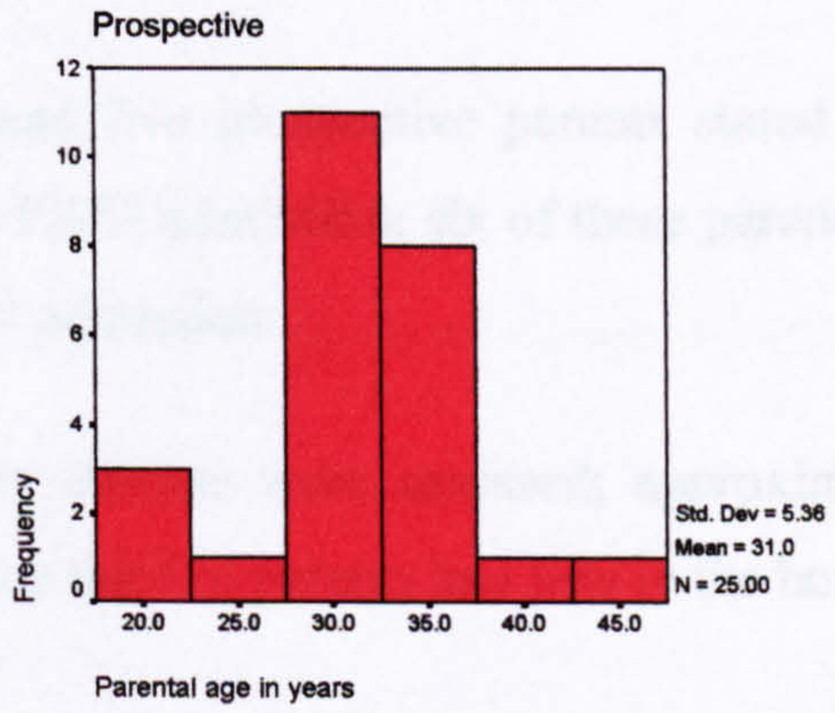


Figure 5.2 Histogram of parental age in years (prospective sample) - Phase II

'Original in colour'

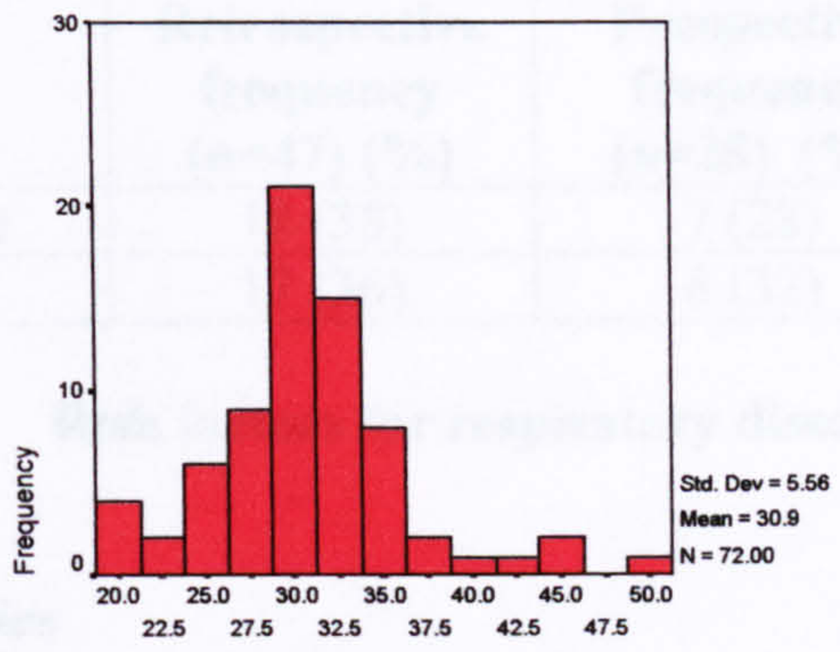


Figure 5.3 Histogram of parental age in years (all parents) - Phase II

Most of the retrospective parents were housewives but there was a large proportion of missing data (Table 5.8). Most prospective parents worked full-time for more than 30 hours per week, or were housewives (Table 5.8).

Employment status	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Full-time > 30 hours per week	4 (9)	10 (40)	14 (19)
Part-time < 30 hours per week	4 (9)	5 (20)	9 (13)
Unemployed not due to illness	0 (0)	1 (4)	1 (1)
Housewife	8 (17)	8 (32)	16 (22)
Maternity leave	0 (0)	1 (4)	1 (1)
Missing data*	31 (66)	0 (0)	31 (43)

*This item was added to the retrospective questionnaires after interview 31.

Table 5.8 Parental employment status (Phase II)

Six retrospective parents and five prospective parents stated that their work status had changed since their child's PICU admission; six of these parents were off work because of their child's health or PICU admission.

Risk factors for respiratory disease were assessed; approximately one third of parents smoked in the home, and one third of parents had pets in the home (Table 5.9).

'Original in colour'

Risk factor	Retrospective frequency (n=47) (%)	Prospective frequency (n=25) (%)	Total (%) (n=72)
Smoking in home	18 (38)	7 (28)	25 (35)
Pets in home	17 (36)	8 (32)	25 (35)

Table 5.9 Risk factors for respiratory disease (Phase II)

5.6.3 Child demographics

Results will be presented separately for retrospective and prospective samples. The mean length of time post PICU discharge for the retrospective sample was 8.7 months. The prospective interviews were conducted 48.0 – 118.5 hours after the child's PICU admission with a mean time of interview of 76.7 hours post PICU admission.

The mean age of children from the retrospective sample was lower than the prospective sample (237.7 days or 7.9 months; SD 278.3 days; range 13-1168 days v. 402.6 days or 13.42 months; SD 467.1 days; range 23-1666 days) (Figures 5.4-5.5). The overall mean age of all children was 294.9 days or 9.8 months (SD 360.8 days; range 13-1666 days) (Figure 5.6). The distribution of age is positively skewed.

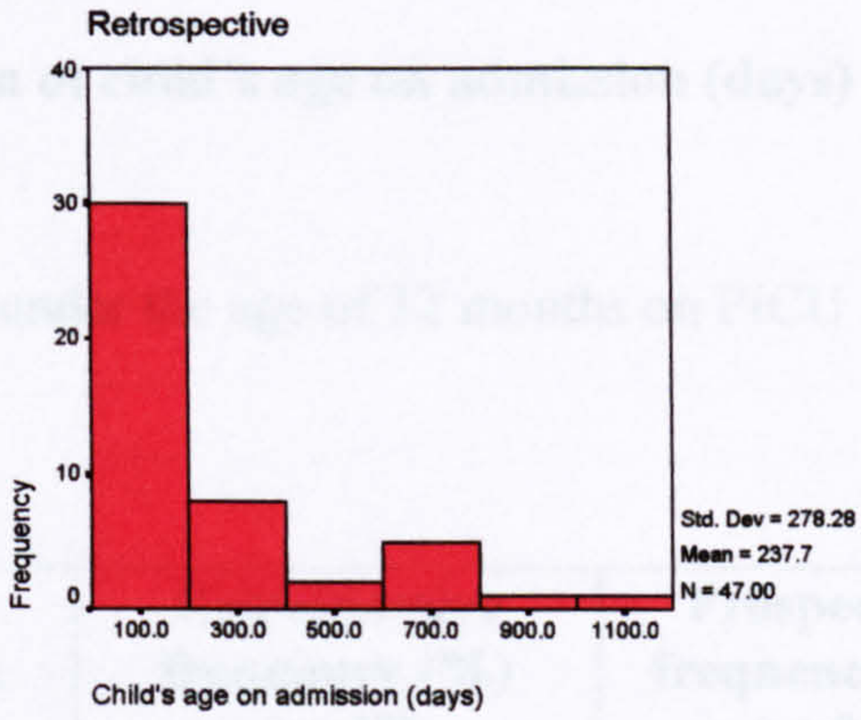


Figure 5.4 Histogram of child's age on admission (days) - retrospective (Phase II)

'Original in colour'

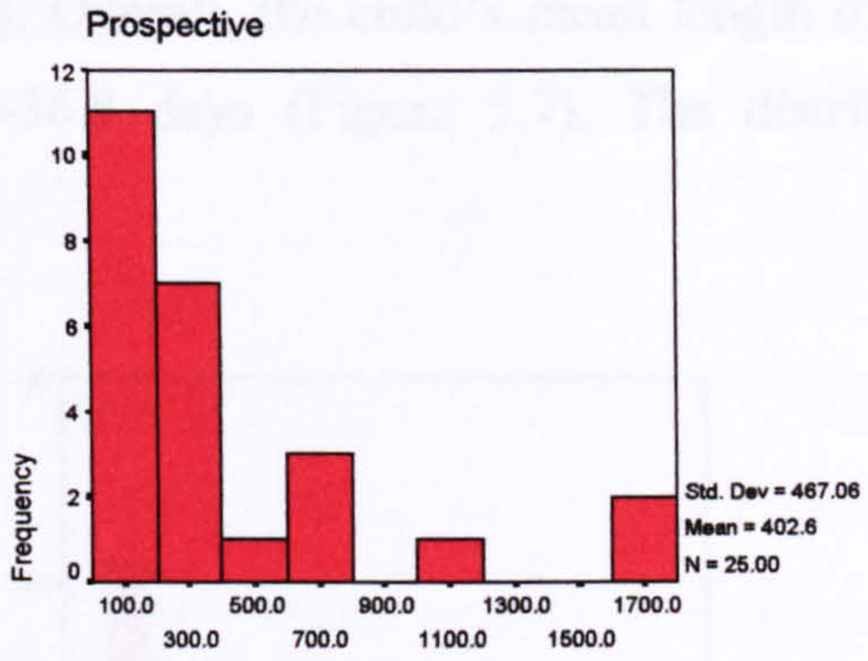


Figure 5.5 Histogram of child's age on admission (days) - prospective (Phase II)

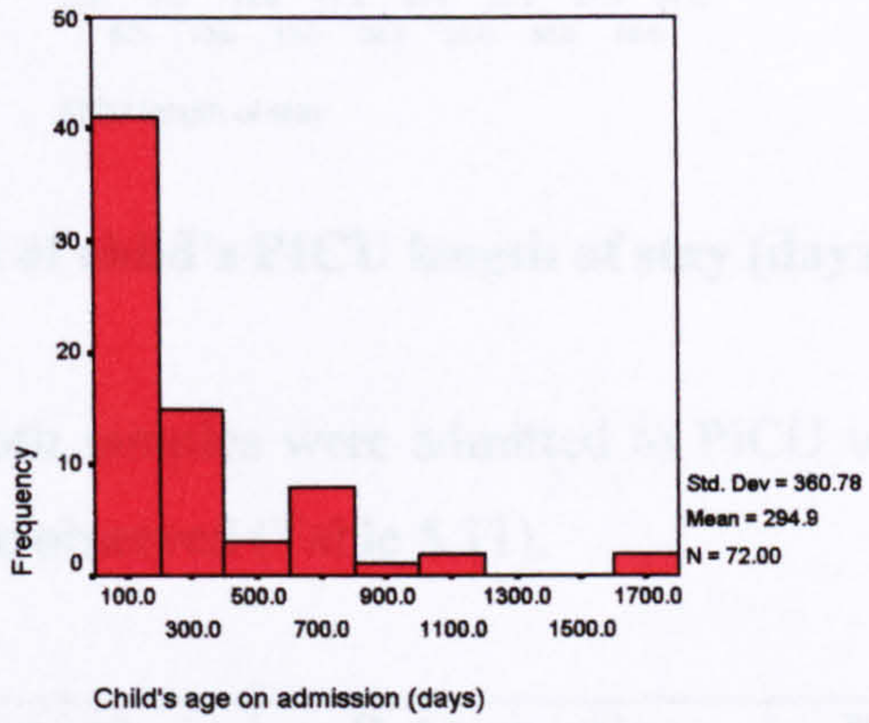


Figure 5.6 Histogram of child's age on admission (days) - all children (Phase II)

Most of the children were under the age of 12 months on PICU admission in both samples (Table 5.10):

Age of child on PICU admission	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
< 12 months	38 (81)	18 (72)	56 (78)
12-23 months	4 (9)	3 (12)	7 (10)
24-35 months	4 (9)	1 (4)	5 (7)
> 36 months	1(2)	3 (12)	4 (6)

Table 5.10 Child's age on PICU admission by sample type (Phase II)

The child's mean PICU length of stay (LOS) was longer for the retrospective sample (10.5 days; SD 7.9 days; range 3.0-36.8 days) than the prospective sample (6.3 days; SD 4.8

days; range 2.0-19.1 days). Overall, the child's mean length of PICU stay was 9.0 days (SD 7.2 days; range 2.0-36.8 days (Figure 5.7). The distribution for PICU LOS is positively skewed.

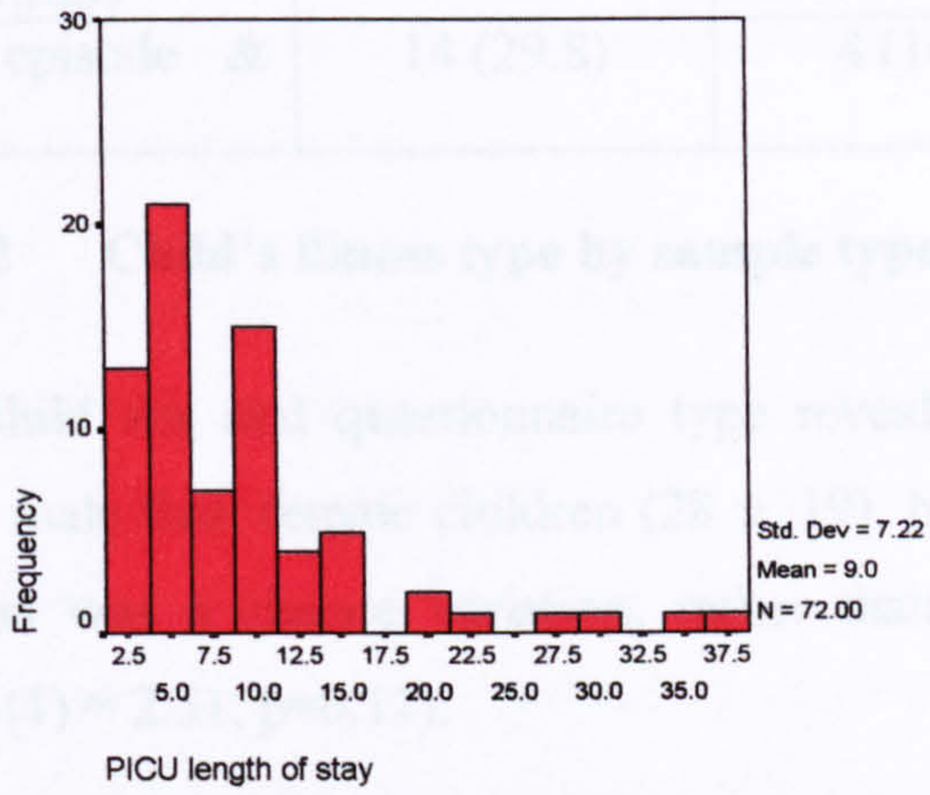


Figure 5.7 Histogram of child's PICU length of stay (days) - all children (Phase II)

Most of the children in both samples were admitted to PICU with bronchiolitis but other respiratory conditions were observed (Table 5.11).

PICU admission diagnosis	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Upper respiratory tract infection (E.g. whooping cough, croup, epiglottitis)	4 (9)	4 (16)	8 (11)
Lower respiratory tract infection (E.g. bronchiolitis, pneumonia)	28 (60)	9 (36)	37 (51)
Respiratory failure	10 (21)	7 (28)	17 (23)
Mechanical airway problem (E.g. subglottic stenosis, tracheal cyst, vocal chord palsy)	1 (2)	4 (16)	5 (7)
Post-operative surgery/problem to airways	4 (9)	1 (4)	5 (7)

Table 5.11 Child's PICU admission diagnosis by sample type (Phase II)

The child's illness type was classified as acute respiratory, acute and chronic respiratory or acute respiratory and other chronic illness. Most children in both samples were admitted with an acute respiratory illness (Table 5.12).

Classification of illness type	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Acute respiratory episode	22 (46.8)	13 (52.0)	35 (48.6)
Acute respiratory episode & chronic respiratory illness	11 (23.4)	8 (32.0)	19 (26.4)
Acute respiratory episode & other chronic illness	14 (29.8)	4 (16.0)	18 (25.0)

Table 5.12 Child's illness type by sample type (Phase II)

A cross tabulation of child sex and questionnaire type revealed that the retrospective sample comprised more male than female children (28 v. 19), but the difference was not statistically different and was a chance variation, rather than a systematic difference between the samples (χ^2 (1) = 2.51, p=0.11).

Most children were first or second born (Table 5.13).

Birth order	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
First	10 (21)	14 (56)	24 (33)
Second	17 (36)	5 (20)	22 (31)
Third	11 (23)	4 (16)	15 (21)
Fourth	6 (13)	0 (0)	6 (8)
Other	3 (6)	2 (8)	5 (7)

Table 5.13 Child's birth order by sample type (Phase II)

Prospective parents were asked if their child was a multiple birth; four children were one of twins and 23 children were singletons. Sixteen retrospective and seven prospective children were born prematurely, with a gestational age ranging from 26-38 weeks and 30-38 weeks respectively. Most children (36/47 = 77% retrospective; 19/25 = 76% prospective) had previously been admitted to hospital prior to PICU admission on 1-5 occasions or more. Four children (16%) from the prospective sample had been admitted to PICU on one occasion previously. The main reason for admission was for respiratory problems (28/36 = 78% retrospective, 12/19 = 63% prospective). A few children were admitted to hospital from birth owing to prematurity (5/36 = 14% retrospective; 3/19 = 16% prospective). Thirty-two retrospective children (68%) were admitted to hospital following PICU discharge on 1-5 occasions or more; one child was an in-patient since

PICU discharge. The main reason for hospital admission following PICU discharge was for respiratory problems (17/32 = 53%) and other non-respiratory illnesses (7/32 = 22%). Thirty-five children (75%) from the retrospective sample, and 13 children (52%) from the prospective sample had not visited a GP for any health reason in the previous month. However, of those children that did visit the GP, twelve visits (100%) were related to a respiratory problem for both sample types.

Twenty-nine children (62%) from the retrospective sample and seven children (28%) from the prospective sample received medicines or treatment at home. Parents described fifty-one drug treatments and nine other treatments (e.g. physiotherapy, specialised feeds, speech therapy, etc.). Eight children (17%) from the retrospective sample and three children (12%) from the prospective sample received oxygen therapy at home day or night (0.0-0.5 litres/minute v. 0.2-0.8 litres/minute). Parents were asked if they had been told by a health care professional if their child had a particular health problem. Twenty-four children (51%) from the retrospective sample and five children (20%) from the prospective sample were reported to have a chronic respiratory problem, such as asthma or cystic fibrosis or chronic lung disease. Other categories of health problem that scored highly were developmental delay (15/47 = 32% retrospective), digestive/gut problems (15/47 =32% retrospective; 5/25 = 20% prospective), heart problems (9/47 = 19% retrospective), learning problems and neurological/brain problems (4/25 = 16% respectively), and speech problems (11/47 = 23% retrospective; 5/25 = 20% prospective).

5.6.4 Practicality

Most parents (50%) found the questionnaire 'quite easy' to complete (Table 5.14).

Ease of completion of questionnaire	Retrospective frequency (%) (n=47)	Prospective frequency (%) (n=25)	Total (%) (n=72)
Very easy	8 (17)	4 (16)	12 (17)
Quite easy	27 (57)	9 (36)	36 (50)
Neither easy nor difficult	7 (15)	5 (20)	12 (17)
Quite difficult	3 (6)	3 (12)	6 (8)
Difficult	1 (2)	2 (8)	3 (4)
Missing data	1 (2)	2 (8)	3 (4)

Table 5.14 Parental questionnaire evaluation (Phase II)

The questionnaire was interview-administered and completion times were 25-90 minutes (mean 46.0 minutes, SD 13.8 minutes) for retrospective parents and 15-60 minutes (mean 35.2 minutes, SD 12.5 minutes) for prospective parents. Overall, the mean completion time was 42.2 minutes (SD 14.2 minutes).

5.6.5 Item reduction

Data from retrospective and prospective interviews have been integrated for the purposes of this analysis. Endorsement frequencies were calculated for all items; items outside the range of 0.2-0.8 were considered for removal from the questionnaire (Streiner & Norman, 1995). Endorsement frequencies for retrospective and prospective response options fell within the acceptable endorsement range (0.2-0.8) for the majority of items, with the exception of seventeen items. Two items in the child demographic section on past medical history (diabetes and chronic rheumatic disease) were removed from the questionnaire as 100% of parents reported that they had not been told by a health care professional that their child had these conditions. A further fourteen items in this section, and one item on oxygen therapy at home, also fell outside the acceptable endorsement range for at least one response option. These fourteen items are rare, with 1-15% of parents not replying 'no' (i.e. recording 'yes' or 'don't know' response options). However, following discussion with clinicians in the RAG, these demographic items although rare, were retained because of their clinical importance.

An item on medication/treatment use fell within the acceptable endorsement range, but parents identified a large number (n=60) of medications and treatments. Large amounts of data were generated from the open-ended question asking parents to list the name, amount and frequency of medications used; this was unwieldy to analyse and required considerable effort from parents to complete the item. Furthermore, the information could be obtained from other data sources, such as the child's GP or hospital record if needed. The item on medication/treatment use and related open-ended question were therefore omitted from the demographic section of the questionnaire used in Phase III (Chapter 6), following discussion with clinical RAG members.

5.6.6 Internal consistency and homogeneity of the HRQoL measure

Corrected item-total correlations were calculated for items in the PICQoL questionnaires Section 1 on daily activities (Qu. 1.4 retrospective/Qu. 1.2 prospective), IPQ (Qu. 1.6

retrospective/Qu. 1.4 prospective), impact on family (Qu. 1.7 prospective/Qu. 1.5 prospective), parental worries about health (Qu. 1.8-1.11 retrospective/Qu. 1.6-1.9 prospective), STAI (Qu. 1.12 retrospective/Qu. 1.10 prospective); and PICQoL questionnaires Section 2 on respiratory symptoms (Qu. 2.3 retrospective/Qu. 2.2 prospective), and respiratory limitations (Qu. 2.4 retrospective/Qu. 2.3 prospective) (Appendices VII-VIII). Results are summarised in Table 5.15. Single items (general health, pre and post PICU health comparison, general breathing, and comparison of pre and post PICU breathing) were not included in the analysis.

Items	Number of items	Number of respondents	Range of corrected item-total correlations
Daily activities	27	72	-0.02-0.72
IPQ minus causal items	16	72	-0.01-0.46
Impact on family life	6	72	0.45-0.84
Worries about health	4	72	0.49-0.77
STAI: Y-6	6	72	0.62-0.76
Respiratory symptoms	18	72	0.04-0.64
Limitations owing to breathing	6	72	0.56-0.82

Table 5.15 Range of corrected ITCs of PICQoL items (Phase II)

All items correlated with the total score above 0.2, with the exception of 'pain, anxiety, sad, happy, angry, cries, tired' and 'quiet' (daily activities); 'short, permanent, long, serious, consequences, easy, economic, improved, symptoms, nothing, treatment, recovery' and 'child' (IPQ); and 'frequency and severity of snuffles' (respiratory symptoms). However, the IPQ items were retained because they have been previously validated, and the other items were retained because they met the endorsement range criterion. In the impact on family life items, the 'stopped you going to work item' was removed as the value for alpha increased markedly if the item was deleted (α=0.91).

The internal consistency was calculated using Cronbach's alpha statistic, with alpha values falling within an acceptable range (0.7-0.9), with the exception of the IPQ minus causal items (α=0.57) and respiratory symptoms items (α=0.92) (Table 5.16).

Question	Number of respondents	Cronbach's alpha statistic
Daily activities	72	0.76
IPQ minus causal items	72	0.57
Impact on family life	72	0.88
Worries about health	72	0.83
STAI: Y-6	72	0.88
Respiratory symptoms	72	0.92
Limitations owing to breathing	72	0.90

Table 5.16 Internal consistency of items (Phase II)

5.6.7 Dimensionality of the HRQoL measure

Conducting FA of the daily activities items assessed dimensionality of the measure in addition to facilitating item reduction. Factor analysis aims to reduce a large number of observed variables to a smaller number of factors to eliminate redundancy, and to identify structure (Tabachnick & Fidell, 2001). Factor analyses of the IPQ items and respiratory symptoms items from Phase II are described in Chapter 6 in relation to the development of a scoring system for the PICQoL questionnaire and reliability of the IPQ.

Factor analysis of the daily activities items was conducted on pooled data (n=72) because the sample size for retrospective data and prospective data alone was too small. Missing values were replaced with the median values for each item. Daily activities items generated for the Phase II questionnaire aimed to measure physical, emotional, social and psychological aspects of a child's health. Principal axial factoring was performed through SPSS 10.0 on 27 daily activities items for a sample of 72 parents, using one-factor, two-factor and three-factor solutions with orthogonal rotation (varimax) and oblique rotation (direct obliminal). The scree plot suggested that there might be two or three factors (Figure 5.8).

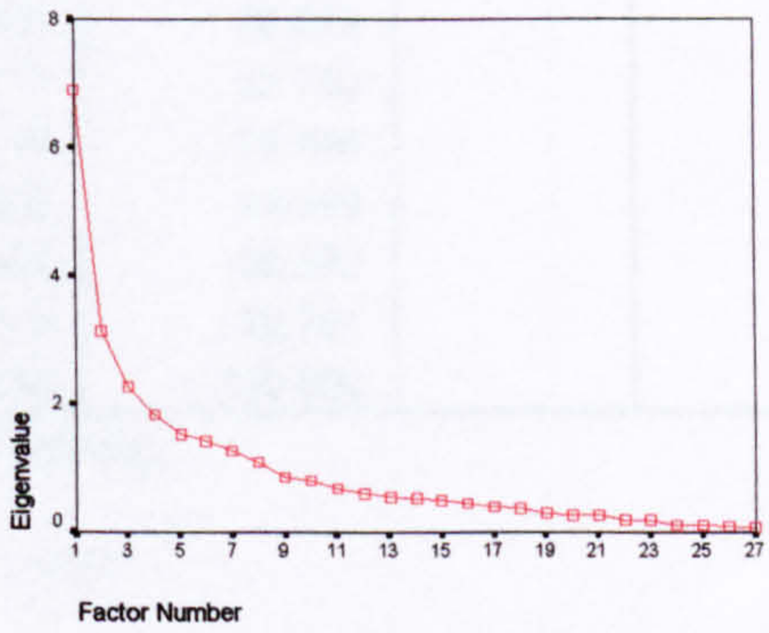


Figure 5.8 Scree plot of PAF daily activities items - Qu. 1.4 - all parents (Phase II)

Results are presented using the approach reported by Bryman and Cramer (2001). With a cut-off factor loading score of 0.3, 25.5% of the variance is explained by one factor (Appendix IX). Nineteen of the 27 items (70%) loaded on one factor, some with skewed distributions (Appendix IX). A cluster of non-loads related to negative emotional state ('plays on own', 'feeling pain', 'anxious', 'behaves', 'quiet', 'angry', tantrums'), suggesting that there may be structure. In the PAF two-factor solution, with direct obliminal rotation, Factors 1 and 2 correlated as – 0.257 (Appendix IX). As the correlation was low, orthogonal (varimax) rotation was selected. In PAF with a two-factor solution, orthogonal (varimax) rotation and 0.3 as a cut-off score, the first factor accounted for 25.52% of the variance and the second factor accounted for 11.69% of the variance (Table 5.17).

Total Variance Explained

Factor	Initial Eigenvalues			Rotation Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	6.890	25.520	25.520	5.541	20.522	20.522
2	3.155	11.686	37.206	3.360	12.444	32.967
3	2.262	8.379	45.585			
4	1.837	6.802	52.387			
5	1.540	5.704	58.092			
6	1.425	5.279	63.371			
7	1.282	4.747	68.118			
8	1.093	4.050	72.168			
9	.859	3.182	75.350			
10	.826	3.061	78.411			
11	.688	2.549	80.960			
12	.623	2.306	83.267			
13	.563	2.086	85.353			
14	.535	1.982	87.335			
15	.509	1.885	89.220			
16	.450	1.667	90.887			
17	.400	1.480	92.367			
18	.373	1.383	93.750			
19	.319	1.181	94.931			
20	.281	1.041	95.972			
21	.270	1.001	96.973			
22	.210	.777	97.750			
23	.198	.734	98.484			
24	.126	.465	98.950			
25	.117	.433	99.382			
26	.102	.379	99.761			
27	6.453E-02	.239	100.000			

Extraction Method: Principal Axis Factoring.

Note: E-02=10^{-2} so 6.453E-02 = 0.06453

Table 5.17 PAF (2-factor solution) variance of daily activity items - all parents (Phase II)

The analysis produced two well-defined factors: a positive developmental milestones factor and a negative emotional factor. In the PAF three-factor solution (varimax rotation), 45.6% of the variance is explained by three factors with a cut-off factor loading score of 0.3 (Appendix IX). A small group of items (excluding double-loaded items) appeared to constitute a third miscellaneous factor, which may be an introverted behaviour factor, but there were too few items to really constitute a clear factor. The first factor described physical milestones and the second factor described negative emotions. Similar factors were also identified in the PAF analysis with a three-factor solution and oblique (direct obliminal) rotation (Appendix IX). The PAF one-factor and three-factor solutions were therefore rejected because of their lack of structure and the two-factor solution accepted. Loadings of items on factors were ordered and grouped by size of loading to facilitate interpretation in the PAF two-factor solution (varimax) rotation (Table 5.18).

Rotated Factor Matrix[a]

	Factor	
	1	2
Holds & carries things - recoded	.889	
Grasps or picks things up - recoded	.785	
Child plays with friends - recoded	.647	
Level of movement - recoded	.646	
Child learns - recoded	.637	-.316
Walks or runs - recoded	.622	
Child's speech - recoded	.602	-.436
Child understands - recoded	.600	-.336
Child plays with siblings - recoded	.582	
Child is naughty - recoded	.566	.350
Child's vision - recoded	.554	
Child's weight - recoded	.318	
Child's hearing - recoded		
Child pays attention - recoded		
Child is angry - recoded		.723
Child cries - recoded		.717
Child is sad - recoded		.534
Child has tantrums - recoded		.497
Child is happy - recoded		-.463
Child feeling pain - recoded		.460
Child laughs - recoded	.398	-.425
Child is anxious - recoded		.371
Child's height - recoded	.315	-.353
Child is tired - recoded		
Child is quiet - recoded		
Child plays on own - recoded		
Child behaves - recoded		

Extraction Method: Principal Axis Factoring.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 3 iterations.

Table 5.18 PAF (2-factor solution) rotated factor matrix of daily activities items – all parents (Phase II)

The rotated component (varimax) matrix produced six double loads: 'learns', 'speech', 'understands', 'naughty', 'laughs' and 'height'. Items were removed if the difference between the double loads was greater than 0.1. 'Learns' loaded more highly on Factor 1 than Factor 2 (0.637 v. –0.316) and so was removed from Factor 2. 'Speech' loaded more highly on Factor 1 than Factor 2 (0.602 v. –0.436) and was therefore removed from Factor 2. 'Understands' loaded more highly on Factor 1 than Factor 2 (0.600 v. –0.336) and was removed from Factor 2. 'Naughty' loaded more highly on Factor 1 than Factor 2 (0.566 v. 0.350) and was removed from Factor 2. Perhaps parents distinguished between 'angry' /'tantrums' etc. and 'naughty' which they viewed in both a positive and negative light. 'Height' was removed from both factors owing to the small difference in the loadings between factors (0.038), it also did not fit conceptually in Factor 2. 'Laughs' was retained in Factor 2 as it loaded more highly on this factor and fitted well conceptually. Twelve items were retained in Factor 1 and eight items in Factor 2. Six items did not load on a factor: 'hearing', 'pays attention', 'tired', 'quiet', 'plays on own' and 'behaves'; these items may be considered for exclusion in future questionnaires.

Factor 1 describes a positive developmental milestones factor ('holds', 'grasps', 'plays with friends', 'moves', 'learns', 'walks', 'speech', 'understands', 'plays with siblings', 'naughty', 'vision', and 'weight'). Factor 2 describes a negative emotions factor ('angry', 'cries', 'sad', 'tantrums', 'happy', 'pain', 'laughs' and 'anxious'). The negative load on 'happy' and 'laughs' strengthens this factor; these items were not reverse coded. Item-total correlations of the factors from the FA of daily activities items were calculated and are summarised (Table 5.19). The alpha coefficient for the positive developmental milestones factor was high (α=0.89) and was low for the negative emotions factor (α=0.44). The alpha coefficient in the first factor suggests that the factor has high internal reliability; the items in the second factor may not however be tapping the same underlying area of interest (Jenkinson & McGee, 1998).

Factor	Number of items	Number of respondents	Range of item-total correlations
Positive developmental milestones	12	72	0.33-0.73
Negative emotions	8	72	-0.22-0.53

Table 5.19 Range of ITCs of factors from FA of daily activity items (Phase II)

5.6.8 Standardised measures (STAI & IPQ)

Mean STAI and IPQ scores are summarised (Tables 5.20-5.21). Mean STAI scores were higher for prospective parents than retrospective parents, and fathers than mothers. T-tests of STAI scores by sample type revealed significant group differences ($p<0.001$), but no significant differences were found between STAI scores of mothers or fathers. Mean IPQ consequences and cure/control scores were higher for prospective parents than retrospective parents; mean IPQ timeline scores were approximately equal in both sample types. Mean IPQ symptom score and timeline scores were higher for mothers than fathers, while mean IPQ consequences and cure/control scores were approximately equal. T-tests of IPQ scores by sample type revealed significant groups differences for cure/control scores ($p<0.05$), but no significant differences were found between IPQ scores of mothers and fathers.

Mean scores (SD)	All parents (n=72)	Retrospective (n=47)	Prospective (n=25)	t-test	
				95% CI for difference	Sig.
STAI score	48.1 (17.9)	39.8 (14.9)	63.7 (66.7)	(-0.3, -17.1)	<0.001
IPQ illness identity (symptom score)	8.9 (3.0)	9.17 (3.1)	8.2 (2.6)	(-0.5, +2.4)	0.21
IPQ timeline	2.6 (0.7)	2.6 (0.7)	2.5 (0.6)	(-0.2, +0.5)	0.41
IPQ consequences	3.1 (0.7)	3.0 (0.7)	3.3 (0.8)	(-0.7, - 0.0)	0.07
IPQ cure/control	3.0 (0.6)	2.8 (0.6)	3.2 (0.5)	(-0.7, -0.1)	0.02

Table 5.20 Mean (SD) IPQ/STAI scores and t-tests by sample type (Phase II)

Mean scores (SD)	Mothers (n=56)	Fathers (n=16)	t-test	
			95% CI for difference	Sig.
STAI score	47.7	49.6	(-8.3, +12.1)	0.71
IPQ illness identity (symptom score)	9.0	8.4	(-2.3, +1.1)	0.48
IPQ timeline	2.6	2.3	(-0.7, +0.1)	0.10
IPQ consequences	3.1	3.2	(-0.2, +0.6)	0.44
IPQ cure/control	2.9	3.1	(-0.2, +0.5)	0.46

Table 5.21 Mean (SD) IPQ/STAI scores and t-tests by parental sex (Phase II)

5.7 Discussion

This phase involved the development of the PICQoL questionnaire and investigated the item selection (reduction), homogeneity (internal consistency), and dimensionality of the measure in a population of parents whose children were admitted to PICU with a respiratory illness. Relationships between parental anxiety and parental illness perceptions were also explored. Piloting the questionnaire in context resulted in the addition of a few HRQoL items in the daily activities section and modifications to the layout of the frequency and severity symptoms section. The IPQ was modified to be more appropriate to childhood illnesses and items were added to the child and parent demographic sections.

The PICQoL questionnaire was designed to include mainly closed questions with open-ended questions used sparingly; general questions preceded specific ones; and demographic questions were placed at the end of the questionnaire in Sections 3 and 4 (McColl et al, 2001). It is recommended that the ordering of generic and disease-specific measures should follow the rules for general versus specific questions (McColl et al, 2001). This HRQoL measure commenced with general questions about the child's health (Section 1), and then progressed to respiratory-specific questions (Section 2). Evidence suggests that question wording and framing, including the choice and order of response categories can have an important impact on the nature and quality of responses (McColl et al, 2001). Although not specifically tested in this phase, parental evaluation of the questionnaire (Section 5) illustrated that most parents (66.6%, n=48) found the questionnaire was 'very easy' or 'quite easy' to complete. However, the mean completion time was relatively long at 42.2 minutes. It must be acknowledged that the questionnaire was administered face-to-face by the author and this is not the intended mode of future administrations; the questionnaire is designed for self-administration. This may impact upon future completion times and response rates (McColl et al, 2001).

Endorsement rates and ITCs were the procedures used for item selection. Endorsement rates for at least one response option per item were within the accepted range (0.2-0.8) for the majority of items, with the exception of a few items relating to the child's past medical history and oxygen use at home (Section 3). These items were retained, as clinicians believed they were relevant to the assessment of a child's HRQoL. It was decided to remove the 'anxiety' item from the daily activities question (Qu. 1.4 retrospective/Qu. 1.2 prospective) in future questionnaires, despite it meeting the endorsement and ITC criteria,

as parents of younger children expressed difficulty in applying the concept of anxiety to their young child. This became evident during the face-to-face administration of the measure post-pilot.

Item-total correlations measure the strength of the correlation between each item and its constituent scale (McColl et al, 2001). Corrected item-total correlations for some PICQoL items were outside the acceptable range of >0.2, but because these items met the endorsement range criterion they were retained. The internal consistency of the items on daily activities, impact on family life, worries about health, STAI and limitations owing to breathing were high with alpha coefficients ranging from 0.76-0.90. However, the internal consistency for the IPQ (minus causal) items was low (α=0.57) and was very high for the respiratory symptom items (α=0.92). If alpha coefficients are too high, it may suggest a high level of item redundancy; this may indicate that some of the items are unnecessary and that the scale as a whole may be too narrow in its scope to have much content validity (Streiner & Norman, 1995). The high level of redundancy may be because severity and frequency seem to be measuring the same thing, and either frequency items or severity items could be left out which would halve the number of respiratory symptom items.

Exploratory FA aimed to describe and summarise the data by grouping together variables that are correlated, and to assess the dimensionality and selection of certain items in the measure (Streiner & Norman, 1995; Kinnear & Gray, 1995; Tabachnick & Fidell, 2001). Ideally, pooling of several samples should be avoided as samples known to be different with respect to some criterion may also have different factors (Tabachnick & Fidell, 2001). Pooling results from diverse groups may also obscure differences rather than illuminate them (Tabachnick & Fidell, 2001). However, if different samples do produce the same factors, pooling them is desirable because of the increase in sample size. Orthogonal (varimax) rotations produce factors that are unrelated or independent of one another with the advantage of their ability to achieve simple structure; however, their disadvantage is that the factors may have been forced to be unrelated whereas in real life they may be related (Bryman & Cramer, 2001). In oblique rotation, factors are correlated, and therefore less artificial than in an orthogonal solution (Bryman & Cramer, 2001). Factor analysis can be used to produce a grouping of attitudes; however when the analysis fails to produce a clear and coherent grouping of attitudes, a researcher may be tempted to squeeze or push

attitudes into spurious groups to which equally spurious names may be attached (Oppenheim, 1992).

The retrospective and prospective samples in this study possessed similar demographic characteristics, with the exception of the timing of PICU admission. It was therefore deemed acceptable to pool the data together for the purposes of FA. As discussed previously (see Section 5.5.5) there are many different recommendations for the appropriate sample size to be used in FA, with little consistency. Tabachnick and Fidell (2001) recommend that as a general rule of thumb it is comforting to have at least 300 cases for FA, and that solutions that have several high loading marker variables (>0.80) do not require such large sample sizes, others suggest 150 cases as sufficient (Guadagnoli & Velicer, 1988). Kline (1994) suggests that in scale development, samples of 100 are quite sufficient where data has been factor-analysed and a clear factor structure produced. Nunnally (1978) recommends a minimum of ten respondents per item (Nunnally, 1978). Data was therefore pooled to increase the sample size, as FA of prospective data alone on 25 parents was considered inappropriate.

Factor analysis of the daily activities items (n=27) revealed two factors: positive milestones and a negative emotional factor. The items in this question were originally developed with physical, emotional, social and psychological (cognitive) dimensions of health in mind. The positive milestones factor encompasses physical, social and cognitive aspects of health, while the emotions factor encompasses negative emotions. Although the FA revealed structure, analysis on data from a larger sample is needed to confirm this structure; no items were removed on the basis of the FA at this stage.

A STAI score of '34-36' indicates a "normal" score (Marteau & Bekker, 1992). Results revealed that the mean STAI score was slightly above the 'normal score' range for retrospective parents but was markedly higher for prospective parents, indicating high levels of parental anxiety. Admission of a child to PICU is a stressful experience for most parents and represents a crisis situation; most PICU admissions are emergencies. This was reflected in the higher STAI scores observed for prospective parents, as expected, over retrospective parents. Future research into possible differences in STAI scores between male and female respondents may be beneficial in terms of identifying coping mechanisms

with implications for future psychological care, but no such differences were found in the sample, perhaps because there were fewer males than females.

Retrospective parents perceived their child's illness to last for a slightly shorter time than did prospective parents. However, prospective parents perceived their child's illness to have more consequences and be less controllable than did retrospective parents. These results may be reflective of the timing at which the IPQ items were completed by parents. Retrospective parents were able to reflect on the whole PICU episode, while prospective parents were only able to reflect on part of the PICU episode as their child was still on the PICU at the time of item completion. Therefore, prospective parents may have been less certain about their child's ability to recover from their PICU illness. Differences between male and female respondents in terms of their illness perceptions warrant future investigation.

The quantitative methods utilised in this phase enabled the research objective to be met, namely the selection of items to include within the measure. Face-to-face administration of the PICQoL questionnaire enhanced the response rate and allowed for the identification of any misunderstandings in item wording (Streiner & Norman, 1995). Although, face-to-face administration to parents was costly in terms of time in this study, the trade-off in data quality was considered a particular strength of the approach in this phase. Utilising a prospective sample enhanced the sample size and reduced bias owing to recall and memory distortions.

Respondents in this study included twelve parents (16.6%) from ethnic minorities. Generally, 30% of all PICU admissions are children from ethnic minorities; the low number of parents from ethnic minorities might be a reflection of the limiting nature of the inclusion criteria of English literacy. Over-sampling for these parents in the sampling frame could have minimised this limitation and achieved a more proportionate sample of parents from ethnic minorities.

5.8. Summary

This study established the item selection, homogeneity, internal consistency and dimensionality of the PICQoL questionnaire in a PICU population. Differences were observed as expected in anxiety scores and IPQ consequences and cure/control scores for retrospective and prospective parents. The retrospective PICQoL questionnaire for testing

in the next phase (Phase III) therefore comprised 66 HRQoL items, and included STAI, IPQ, demographic and questionnaire evaluation items. Future prospective versions of the questionnaire will comprise 64 HRQoL items and include the STAI, IPQ, demographic and questionnaire evaluation items. However, the prospective version was not required for Phase III.

CHAPTER 6 – Application of the HRQoL measure in a PIC setting

6.0 Introduction

This chapter describes a quantitative study (Phase III) to test the psychometric properties of the PICQoL questionnaire developed in Chapter 5, in a PICU population. The face and content validity of the measure were previously found to be satisfactory (Chapters 3-4). In order to develop a validated measure, criterion validity, construct validity, internal reliability, test-retest reliability and practicality of the measure need to be assessed (Streiner & Norman, 1995). The research in this chapter aimed to assess the reliability, validity and practicality of the HRQoL measure.

6.1 Background

The purpose of the HRQoL measure is to evaluate the longitudinal HRQoL outcomes for children under the age of five years, following their discharge from a PICU with a respiratory illness. The system-specific measure is designed for clinical application and to be sensitive to change following treatment (MacKeigan & Pathak, 1992; McDowell & Newell, 1996).

As discussed previously (Chapter 1), intensive care medicine has made important contributions to the survival of critically ill patients. However, intensive care requires expensive equipment, a large staff and a large amount of scarce resources (Gemke, 1999; Eiser & Morse, 2001a). There are also important quality assurance issues that have to be resolved involving equity of access to intensive care in different regions, how the distance to a PICU may affect referral patterns, and the social and financial impact on families who have to travel long distances to receive services (Eiser & Morse, 2001a). Traditionally the assessment of critical care has focused largely on mortality and other objective measures of health.

The results of the systematic review (Chapter 2) revealed no measure in this area. However, the HUI measure was identified as the best available 'gold standard' measure for the assessment of criterion validity of the PICQoL questionnaire. However, there are several limitations to the HUI measure as described previously (Chapter 1-2).

The assessment of criterion validity refers to the ability of a measure to correspond with other measures held up as 'gold standards' (Jenkinson & McGee, 1998). In practice, few

studies can truly claim to have evaluated criterion validity, as 'gold standards' are hard to find in this area of research. Exception to this is the use of a shorter form of a measure, e.g. the SF-36 and SF-12 (Jenkinson et al, 1997). The 'gold standard' may be the longer form measure. Criterion validity asks whether health assessment scores are systematically related to an objective criterion (Landgraf & Abetz, 1996). Criterion validity is usually divided into two types: concurrent validity and predictive validity (Streiner & Norman, 1995). Concurrent validity is where the results from two measures administered at the same time are compared (Streiner & Norman, 1995; Pham & Klaassen, 2000). Predictive validity is where the results of a measure predict a future event or outcome (Jenkinson & McGee, 1998). Concurrent validity was assessed in this study by administering the PICQoL and HUI measures together and calculating correlation statistics (Pearson's product moment and Spearman's Rho correlations) of the PICQoL scores and HUI scores (Streiner & Norman, 1995). Moderate correlations were expected. In the absence of a real 'gold standard' measure, construct validity was also assessed.

6.1.1 Construct validity

Construct validity is concerned with the patterns of relations of an HRQoL measure with other more established measures (Cronbach & Meehl, 1955; Fitzpatrick et al, 1992). This may involve examining the extent of agreement of HRQoL scores with laboratory or clinical measures of severity of disease (Guyatt, Walter & Norman, 1987), or the ability of the measure to distinguish between patient groups considered to have different health states (Stewart et al, 1989). Construct validity seeks to provide empirical evidence by asking whether the hypothesised set of questions used to operationalize a given health construct (e.g. physical functioning) inadvertently elicits information about other independent constructs (e.g. self-esteem) (Landgraf & Abetz, 1996). Hypotheses are generated and the questionnaire is tested to determine if it actually reflects these prior hypotheses (Jenkinson & McGee, 1998). Usually the hypothesis will explore the difference between two or more populations who would be expected to have differing amounts of the property assessed by the measure (Streiner & Norman, 1995). Applying the measure to the appropriate samples tests the hypothetical construct. If the expected relationship is found, then the hypothesis and the measure are sound; conversely, if no relationship is found, the fault may lie with either the measure or the hypothesis (Streiner & Norman, 1995).

Convergent and discriminative validity are two forms of construct validity. For convergent validity to exist, the results from a measure would be related to other variables and measures of the same construct. Discriminant validity assumes that the results will not be related when the questionnaire data are compared to other data measuring distinct and unrelated concepts (Jenkinson & McGee, 1998). Construct validity was tested in this study by exploring the statistical relationship between the child's illness type and PICQoL scores. This analysis aimed to test differences in parental anxiety, parental perceptions of illness, and illness type. Hypotheses were generated from evidence in the literature on parental anxiety in a PICU setting, and parental illness perceptions of healthy and chronically ill children. The hypotheses were theoretically driven as they were informed by the analysis of parental data in Phase I (Chapter 3), and they were also clinically driven as they were informed by the clinical expertise of the author.

Evidence in the literature suggests that a child's admission to a PICU is stressful for parents who feel overwhelmed, helpless and anxious by the unfamiliarity of the environment (Cox, 1992; Hazinski, 1992; Fisher, 1994). Parents also have a decreased ability to think clearly and to problem solve and reduced ability to utilise incoming information in this situation (Rennick, 1986; Hazinski, 1992). This type of iatrogenic consequence is usually assessed using a measure of anxiety. Several measures exist for use in hospital or outside hospital and are described previously (Chapter 5); the short-form STAI was used in this study. Medical knowledge and/or experience with illness may also influence raters' valuations of health states (Froberg & Kane, 1989). The measurement of HRQoL in children is influenced by other factors such as child and parent health and illness cognitions, age, cognitive development, sex, parental social status and non-specific or placebo effects (Froberg & Kane, 1989) (Chapter 3). Evidence also suggests that patient's representations determine coping behaviours, which in turn lead to the outcomes observed (Johnston et al, 1990). In a study of parents of children with chronic illnesses (asthma, diabetes, epilepsy) and parents of children with no chronic illness, the parents of children with chronic disease perceived their own child's disease to be less serious than did doctors or other parents (Marteau & Johnston, 1986). The results suggested that parents' representations of their children's conditions were changed by their experience of the condition from the time of onset (Johnston et al, 1990). These perceptions may be as a result of parental experience of their child's disease and the way in which this experience

is processed cognitively; social processes, including communications between patients and health care staff, may also influence representations (Johnston et al, 1990).

In consequence, two constructs exist: parental anxiety as an outcome measure and parental illness perceptions as a process measure. The following hypotheses were tested utilising these constructs:

- Differences exist in parental anxiety according to the child's illness type
- Differences exist in parental illness perceptions according to the child's illness type
- Differences exist in parental perceptions of child's general health and wellbeing according to the child's illness type
- Differences exist in parental perceptions of child's respiratory health and wellbeing according to the child's illness type
- Differences exist in parental perceptions of the impact of a child's illness on family life according to the child's illness type.

It was hypothesised that parents whose children were admitted to a PICU with an acute respiratory illness would have greater levels of anxiety about their child's health and wellbeing than those parents whose child also had a chronic respiratory illness. It was also hypothesised that differences in parental illness perceptions would be observed for parents of children with acute versus chronic respiratory illnesses, and this may consequently be reflected in differences in the child's HRQoL. It was hypothesised that parents of children admitted to a PICU with an acute respiratory illness would perceive their child's illness to last for a shorter time than those parents of children who were also admitted with an underlying chronic respiratory illness. Also, parents of acutely ill children would perceive their child's illness to be more controllable and curable than for parents of chronically ill children. However, parents of chronically ill children would perceive their child's illness to have more consequences on their child's life and family life than parents of acutely ill children. Illness representations may also differ between mothers and fathers.

6.1.2 Reliability

Reliability of a measure is its ability to produce the same results under the same conditions; generally assessment of two types of reliability is emphasised in scale development. Test-retest reliability examines if results are produced consistently from the same respondents at different times when there is no evidence that change exists

(Jenkinson & McGee, 1998). It is reported that it may be difficult to practically distinguish measurement error from real changes in QoL in the assessment of test-retest reliability (Fitzpatrick et al, 1992). A second type of reliability measurement is the examination of internal reliability/consistency, the degree of agreement of items addressing equivalent concepts. Inter-rater reliability is also reported for interview-based assessments. The homogeneity of a scale is assessed through ITCs; items should be moderately correlated with each other, and each should correlate with the total score. Internal consistency and homogeneity, and test-retest reliability were the forms of reliability evaluated in this study (see Glossary for definitions of types of reliability).

6.2 Methods

6.2.1 Design

A cross-sectional survey design was employed to facilitate the generalisation of findings from a sample to a population (Fowler, 1993; Creswell, 1994). Data was collected in retrospect of the PICU incident. A retrospective approach was chosen because of the advantage of speed within the limited timescales, and to allow for the results of the study to be available as soon as the data is collected and analysed (Moser & Kalton, 1996). However, disadvantages include a real danger of bias for memory distortions. These may have been minimised by employing a prospective design. However, 114-132 children under the age of five years with a respiratory illness are admitted to the larger PICU per year, including duplicate admissions and deaths, which represents 28-30% of all PICU admissions per year. Similar proportions are reported elsewhere (Segedin, 1999). To achieve a sample size of 100-200 children for this study may have taken a year or more to complete prospectively and was not feasible within the study time constraints.

6.2.2 Sample selection

The value of cross-sectional designs depends crucially on choosing a representative, non-biased sample (Robson, 1995). Confidence in overall results is also dependent on the quality of individual responses (Robson, 1995). Children were selected according to the following inclusion criteria:

- Child was alive at time of sampling
- Child had a primary PICU admission diagnosis of a respiratory illness
- Child was <5 years of age

- Child had a PICU LOS ≥ 2 days
- Child was discharged from PICU between January 1999 and February 2001 (more than two months previously)
- Child had parents who were English literate

Parents of children discharged from PICU were selected from two sites, representing the same NHS Trust (Chapter 3). The larger PICU admits approximately 450 children per year, the smaller unit admits approximately 100 children per year. Children admitted to both units were sampled to enhance the recruitment rate. The total population was used to represent the population under study, namely children under the age of five years admitted to a PICU with a primary admission diagnosis of a respiratory illness. The sampling frame used was the PICU computerised database in each unit; the same conditions for sample selection applied as in Phase I (Chapter 3).

6.2.3 Sample size calculation

The primary objective in this study was to develop the measure (scale) rather than estimate values or compare groups using the measure. For this reason the sample size calculated was based upon previous evidence on scale development where samples of 100-200 were used, rather than a power calculation (Kline, 1986; Guyatt et al, 1993; Kline 1994). Power calculations are routinely used to calculate sample sizes in studies that aim to measure effectiveness of treatments or therapies, such as in clinical trials. They can also be used in non-experimental designs, such as cohort, cross-sectional and case-control studies to determine sample size or to determine how wide confidence intervals will be for a given sample (Bland 1997). For example, in a study comparing two subgroups of a population, power calculations may determine what proportion of the population needs to be sampled; sample size calculations using confidence intervals can also apply to surveys attempting to estimate proportions or means in one group (Bland, 1997).

6.3 Materials

Data was elicited using self-administered postal questionnaires as they are less costly than interviews and allow for a larger sample over a wider geographical area to be targeted (Robson, 1995). Postal questionnaires are useful for investigating sensitive topics, such as HRQoL, as there is more anonymity and so frankness is encouraged in the response (Robson,

1995; Bowling, 1997b). There is also a growing body of evidence that people are more willing to report socially undesirable facts about themselves in self-administered form than in an interview (Fowler, 1996). Poor quality interviewing can also be a significant source of error (Fowler, 1996). Self-administration of questionnaires in this study also allowed respondents to have more time to think carefully about their answers, and data collection and analysis was relatively quick (Fowler, 1993; Robson, 1995; Russell, 1998).

Including mainly five-point response options and some open-ended questions in the questionnaire aimed to provide sufficiently comprehensive response options to fully represent the views of the respondents. However, it has been reported that the pre-coding of response choices may force some respondents to choose inappropriate responses (Bowling, 1997b). Well-worded questions with clear and unambiguous instructions were used in order to obtain valid and reliable responses to the questionnaire, which is reported to be a time-consuming science (Polgar & Thomas, 1998). The pilot testing of the questionnaire in this research programme ensured clarity (Chapter 5). It was not possible to guarantee that the intended respondent completed the questionnaire alone, or to check the honesty or seriousness of responses (Robson, 1995; Russell, 1998). The materials used in this survey were a questionnaire pack incorporating the PICQoL questionnaire (Appendix X), which also incorporated the STAI (short-form) and the Carer-version of the IPQ, and the adapted HUI measure.

6.4 Procedure

General practitioners of selected children were contacted to confirm that a child was alive and well before the mother and father of eligible children were invited by letter to complete and return a PICQoL and HUI questionnaire simultaneously. A stamped-addressed envelope was enclosed with the questionnaire pack; higher response rates are generally demonstrated for stamped mail in comparison with franked or reply-paid envelopes (McColl et al, 1998). A covering letter also provided information about the research study, and follow-up was offered to parents by inviting them to obtain a summary of the research results.

Administering the HUI measure aimed to assess the criterion validity of the PICQoL questionnaire. Administering the PICQoL and HUI questionnaires for a second time, 2-3 weeks from the first, to all those parents who responded, aimed to assess the test-retest

reliability of the measures. To reduce attrition rates, non-responders to the postal questionnaires were sent a maximum of two reminders at 2-3 week intervals (Bowling, 1997b). Information on non-responders was collated in terms of the variables identified in the sampling frame (Moser & Kalton, 1996). The demographic section (Qu. 3.1-3.16, and Qu. 4.2-4.9) of the PICQoL questionnaire was removed for repeat questionnaires (Time 2).

The HUI measure was used in this study to assess criterion validity of the PICQoL questionnaire, and as a pilot for a much larger MRC funded study (UK PICOS). The HUI was slightly modified to create an additional response option, "Question not appropriate to the developmental age of my child", which aimed to assess questions that parents had difficulty completing to inform the version used in the UK PICOS. A revised version of the HUI was piloted in UK PICOS, which included additional explanations for each question on the facing page in the questionnaire. Modifications were made, as the face and content validity of the HUI measure were considered poor. Some of the questions lacked clarity to parents of young children; e.g. *"Which one of the following best describes your child's ability, during the past week, to see well enough to read ordinary newsprint?"* It may be argued that this question is assessing cognition as well as vision, and is not appropriate to a child who is too young to read. A more appropriate question might be, *"Which one of the following best describes your child's ability, during the past week, to see a thin piece of ribbon in front of them?"* Modifying a measure may however affect its overall validity (Cheater, 1998).

6.5 Data analysis

6.5.1 Accuracy of data entry

The reliability of the author's data entry was checked by the PICU audit clerk, for 10% of those PICQoL and HUI questionnaires returned in each timeframe (Time 1 and Time 2). The author resolved any differences. A 10% sample of the entered data was also cross-referenced against the original questionnaires (sample edit). Omission and consistency editing checked for missing data and the consistency of responses, so encouraging confidence in the final results.

6.5.2 Sample representativeness

It was not possible to study the whole UK population of children under the age of five years admitted to a PICU with a respiratory illness; a total sample of children under the age

of five years admitted to a regional PICU with a respiratory illness was therefore chosen to be representative of this population. However, it might be argued that a regional sample is not representative of all of the UK as parents might vary by region in their understanding or interpretation of a question, in their behaviour, or their beliefs about health and illness.

6.5.3 Missing data

The data was explored for patterns of missing values as non-randomly missing values can affect the generalisability of results (Tabachnick & Fidell, 2001). Missing data was substituted by the median value for items in the FA of daily activity items (Qu. 1.4) and respiratory symptom items (Qu. 2.3), and by the mean value for items in the FA of IPQ items (Qu. 1.6). Median and mean values were used to substitute missing data if less than 50% of items were missing per case. These choices were based upon the prior knowledge of the author and observation of the distribution of responses (Tabachnick & Fidell, 2001). Data analysis was also repeated using complete cases to compare with the analysis of cases with estimated missing values (medians or means) to identify any differences.

Data on children of non-responders and responders were compared in respect of the child's age, PICU LOS, and illness type, using t-tests and Chi-square statistics. An ANOVA of child's age and PICU LOS by mailshot response were also calculated. Weightings can be used in statistical analysis to compensate for non-response (Bowling, 1997b).

6.5.4 Item reduction

The frequency of endorsement was calculated for all questionnaire items using the same conditions applied previously (see Section 5.5.3). In practice, only items with endorsement rates for response options between 0.20 and 0.80 were used (Streiner & Norman, 1995). Exploratory FA was also used, as in Phase II, to aid the construction of the measure in terms of identifying its structure and redundant items, thus informing the development of the scoring system for the PICQoL questionnaire. Factor analysis was performed on those questions within the PICQoL questionnaire that comprised many items, namely Qu. 1.4 (daily activities) comprising 26 items and Qu. 2.3 (respiratory symptoms) comprising 20 items. The FA was performed on merged data from Phase II and Phase III, with duplicate parents removed, to increase the sample size for analysis. As in Phase II (see Section 5.5.4), ITCs were calculated for items on daily activities (Qu.1.4), IPQ minus causal items (Qu. 1.6), impact on family life (Qu. 1.7), worries about health (Qu. 1.8-1.11), STAI (Qu.

1.12), respiratory symptoms (Qu. 2.3) and limitations owing to breathing (Qu. 2.4), to test the homogeneity of the scale (Appendix X).

6.5.5 Development of a scoring system

The PICQoL questionnaire comprises single items and multiple items; a scoring system was developed to reflect these differences. A scoring system that calculated scores for each dimension was devised rather than an overall score, as the items measured different attributes and not the same trait (Streiner & Norman, 1995). When there are at least 40 items in a scale, differential weighting contributes relatively little, except added complexity for the scorer (Streiner & Norman, 1995). With fewer than 40 items, or 20 items according to Nunnally (1970), weighting may have some effect and increase the predictive ability of the scale (Streiner & Norman, 1995). Differential weighting was used to calculate scores from items in Qu. 1.4 (k=26) and Qu. 2.3 (k=20) (Note: k=number of items).

Factor analysis of PICQoL questions with less than 40 items was conducted to reduce a large number of observed variables to a smaller number of factors – redundancy, and to identify structure to facilitate the calculation of a score (Tabachnick & Fidell, 2001). A review of the scoring systems of measures identified in the systematic review (Chapter 2), particularly the CHQ (Landgraf et al, 1996), and advice from a statistician and experimental psychologist also informed the development of the scoring system for the HRQoL measure.

6.5.6 Reliability

The reliability of the HRQoL measure was assessed using internal consistency and homogeneity, and test-retest reliability checks. The reliability of the Carer-IPQ was also tested by replicating the FA of the original scale developers using merged Phase II/III data for 16 IPQ items (timeline, consequences and cure/control). Results are reported in Section 6.7.

6.5.6.1 Internal consistency (reliability) and homogeneity

Internal consistency (reliability) of the PICQoL items was measured using the Cronbach's alpha statistic, as described previously (Chapter 5). Ideally, alpha statistics should be above 0.70 (Nunnally, 1978), but probably not higher than 0.90 (Streiner & Norman, 1995).

Good internal consistency results do not necessarily mean that a measure is reliable over time; test-retest reliability will assess this.

An item should correlate with the total scale score above 0.20; items with lower correlations should be discarded (Kline, 1986; Streiner & Norman, 1995). Item-total correlations were also performed on the factors identified from the FA of Qu. 1.4, 2.3 and 1.6 (IPQ).

6.5.6.2 Test-retest reliability

Test-retest reliability of the PICQoL and HUI measures was assessed at an interval of 2-3 weeks. Ideally, a time interval sufficiently short to assume that the underlying process is unlikely to have changed needs to be selected; expert opinion varies from one hour to a year depending on the task, but generally a retest interval of 2-14 days is usual (Streiner & Norman, 1995). Correlation statistics using the Pearson's product moment correlation statistics (or Spearman's rho correlation) were calculated between Time 1 and Time 2 scores for the PICQoL and HUI questionnaires.

6.5.7 Practicality

Measures of HRQoL need to be practical, that is, useful in the clinical setting, easy to use and to interpret; the mode of administration also affects its practicality. Practicality was assessed as in Phase II (see Chapter 5).

6.6 Results 1

6.6.1 Accuracy of data entry

The proportion of agreement was calculated on the double data entry of a 10% random sample of PICQoL and HUI Time 1 questionnaires (n=12) and PICQoL and HUI Time 2 questionnaires (n=5), using Cohen's Kappa statistic (Cohen, 1960):

$$\frac{\text{Number of agreements}}{\text{Number of agreements} + \text{Number of disagreements}}$$

(Cohen, 1960)

The results of the proportion of agreement are displayed (Table 6.1). There were six errors in the PICQoL Time 1 data entry, which were resolved by discussion.

Questionnaire	Proportion of agreement
PICQoL Time 1	0.997
PICQoL Time 2	1.000
HUI Time 1	1.000
HUI Time 2	1.000

Table 6.1 Reliability of data entry (Phase III)

6.6.2 Sample representativeness

Utilising the afore-mentioned entry criteria, 199 PICU admissions were identified from two sampling frames (1st January 1999 - 13th February 2001). However, this number was further reduced (Table 6.2).

Reason for exclusion from sampling frame	Number of admissions
Children died during or post PICU discharge	17
Duplicate elements*	35
Unable to verify child's health status	11
Information on health status obtained too late	4
GP refused to give information on health status	1
Total	**68**

*Representing 18 children.

Table 6.2 Reason for exclusion from the sampling frame (Phase III)

With the above exclusions, a total sample of parents of 131 children were eligible to be sampled. The number of eligible respondents decreased further to 122 mothers (or 244 parents including mothers and their partners) (Table 6.3).

Reason for exclusion	Number of mothers (including partners)
Mothers did not read English	2 (4)
Returned mail from Post Office	6 (12)
Child >5 years of age on questionnaire completion	1 (2)
Total	**9 (18)**

Table 6.3 Reason for further respondent exclusion (Phase III)

One mother expressed difficulty completing the questionnaire, as her child was only three weeks old on PICU admission. A further two mothers did not wish to take part; one mother believed her child's illness was not relevant to the study. These three mothers (and their

partners) were included in the total sample size. Eleven mothers stated that they were single and not living with a partner, these mothers were assumed therefore not to have partners and the response rate for both parents was re-calculated to reflect this (n=233). The response rate for one parent (known as at least one parent) or both parents (known as all parents) completing the PICQoL and HUI questionnaires at Time 1 and Time 2 are described (Table 6.4), with numbers of respondents for Time 1 and Time 2 (Table 6.5).

Questionnaire	At least one parent (%)	All parents (%)
PICQoL Time 1	81/122 (66.4)	121/233 (51.9)
PICQoL Time 2	38/81 (46.9)	55/121 (45.5)
HUI Time 1	76/122 (62.2)	114/233 (48.9)
HUI Time 2	36/76 (47.4)	53/76 (69.7)

Table 6.4 Response rates to postal survey (Phase III)

Questionnaire	Mother/partner pair	Mother alone	Father alone
PICQoL Time 1	40	40	1
PICQoL Time 2	17	20	1
HUI Time 1	38	36	2
HUI Time 2	17	18	1

Table 6.5 Numbers who responded for Time 1 and Time 2 (Phase III)

There were 81 (66.4%) responses to the PICQoL questionnaire from at least one parent in Time 1; 42 (51.9%) responded after the first mailshot, 26 (32.1%) after the second mailshot, and 13 (16.0%) after the third mailshot. Some respondents from the postal survey in Phase III also participated in earlier phases - developing the items (Phase I) and testing the items (Phase II) (Table 6.6). Data from these duplicate parents was removed in the FA of merged Phase II/ III data.

Phase	Duplicate children in Phase III (%)
Phase I	6 (7.4)
Phase II	31 (38.3)

Table 6.6 Number of duplicate children whose parents participated in Phase III

6.6.3 Non-responders and responders

There were 121 responders to the PICQoL questionnaire (Time 1), representing 81 children; there were 82 non-responders representing 41 children. Data on group differences of children of non-responders and responders, and mailshot responses (Time 1 and/or Time 2) by child's age on PICU admission and PICU LOS were compared using t-tests (Tables 6.7-6.8).

Means (SD)	Responders by child ID (n=81)	Non-responders by child ID (n=41)	t-test	
			95% CI for difference	Sig.
Child's age on PICU admission (days)	294.4 (366.4)	212.0 (267.5)	(-45.3, +210.3)	0.20
Child's PICU length of stay (days)	8.6 (7.2)	8.0 (5.1)	(-1.9, +3.1)	0.64

Table 6.7 Means (SD) child's age/PICU LOS and t-tests by respondent type (Phase III)

Means (SD)	Time 1 responders by child ID (n=44)	Time 1 & 2 responders by child ID (n=37)	t-test	
			95% CI for difference	Sig.
Child's age on PICU admission (days)	296.1 (338.4)	292.4 (402.0)	(-167.4, +160.0)	0.96
Child's PICU length of stay (days)	8.8 (7.8)	8.4 (6.7)	(-3.6, +2.9)	0.83

Table 6.8 Means (SD) child's age/PICU LOS and t-tests by mailshot response (Phase III)

No significant differences were found between the child's age on PICU admission and child's PICU length of stay of responders or non-responders, or responders in one or both timeframes (Time 1 and/or Time 2). One-way ANOVA of child's age on PICU admission and child's PICU LOS by mailshot response (1st, 2nd, 3rd or no response) revealed no significant group differences by child's age ($F=1.8$, $df=3,118$, $P=0.15$) or PICU LOS ($F=0.6$, $df=3,118$, $P=0.61$).

'Original in colour'

6.6.4 Child demographics

The mean age of the children (n=81) admitted to PICU was 294.4 days/ 9.8 months (SD 366.4 days/12.2 months; range 12-1666 days/ 1-56 months) (Figure 6.1); the distribution of age is positively skewed.

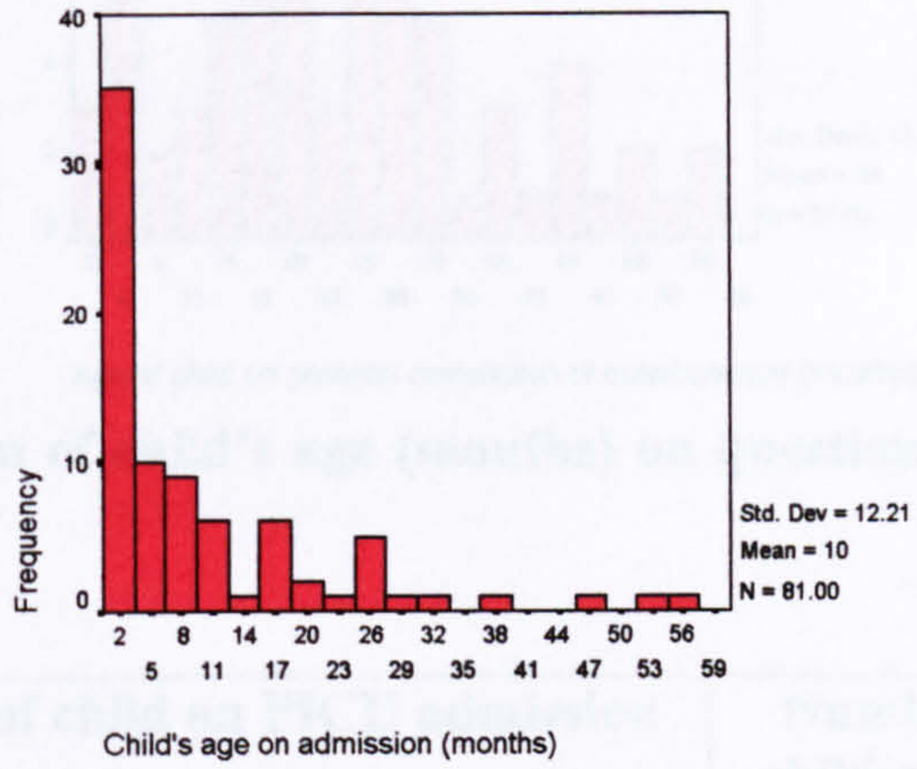


Figure 6.1 Histogram of child's age on admission (months) – Phase III

Sixty (74.1%) children were under twelve months of age on PICU admission (Table 6.9).

Age of child on PICU admission	Number (%)
< 12 months	60 (74.1)
12-23 months	10 (12.3)
24-35 months	7 (8.6)
36-47 months	1 (1.2)
48-60 months	3 (3.7)
Total	**81 (99.9)**

Table 6.9 Child's age on PICU admission (Phase III)

The mean age of the children (n=81) on parental completion of the PICQoL questionnaires was 22.0 months (SD 13.6 months); range 2-56 months (Figure 6.2); the distribution of age is positively skewed. Most of the children (n= 67) were aged less than three years when their parents completed the questionnaires (Table 6.10).

'Original in colour'

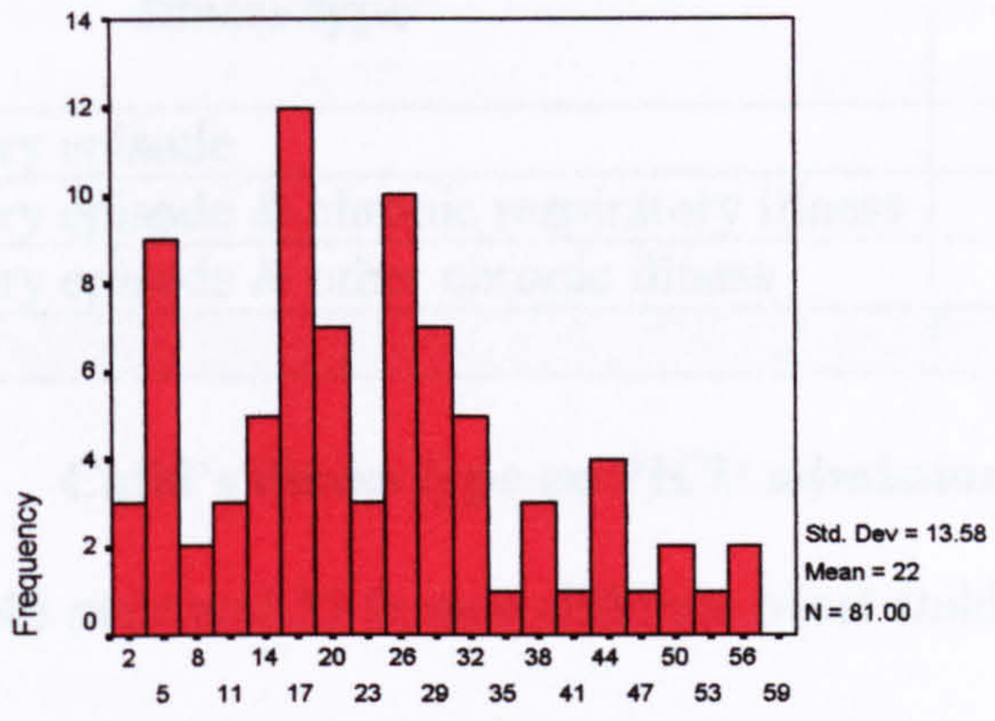


Figure 6.2 Histogram of child's age (months) on questionnaire completion (Phase III)

Age of child on PICU admission	Number of children (%)
< 12 months	18 (22.2)
12-23 months	26 (32.1)
24-35 months	23 (28.4)
36-47 months	9 (11.1)
48-60 months	5 (6.2)
Total	**81 (100.0)**

Table 6.10 Frequencies of child age on questionnaire completion (Phase III)

The child's mean length of PICU stay was 8.6 days (SD 7.2 days, range 2.0-36.8 days). Parents completed the questionnaires 1-26 months after their child's discharge from PICU (mean 12.7 months, SD 8.14 months). The children were admitted to the PICU from 1st January 1999 to 4th February 2001. Most of the children were admitted with bronchiolitis (n=33), but other respiratory conditions were observed (Table 6.11).

PICU admission diagnosis	Number of children (%)
Upper respiratory tract infection (e.g. croup, epiglottitis)	9 (11.1)
Lower respiratory tract infection (e.g. bronchiolitis, pneumonia)	47 (58.0)
Respiratory failure	13 (16.0)
Mechanical airway problem (e.g. vocal chord palsy, obstructive sleep apnoea)	5 (6.2)
Post-operative surgery to airways	7 (8.6)
Total	**81 (99.9)**

Table 6.11 Child's PICU admission diagnosis (Phase III)

The child's illness type was classified using the afore-mentioned criteria (Table 6.12).

Illness type	Number of children (%)
Acute respiratory episode	41 (50.6)
Acute respiratory episode & chronic respiratory illness	13 (16.0)
Acute respiratory episode & other chronic illness	27 (33.3)
Total	**81 (99.9)**

Table 6.12 Child's illness type on PICU admission (Phase III)

The sample comprised 46 male and 35 female children. Most children were first or second born (Table 6.13).

Birth order	Number of children (%)
First	24 (29.6)
Second	26 (32.1)
Third	19 (23.5)
Fourth or more	10 (12.3)
Missing data	2 (2.5)
Total	**81 (100.0)**

Table 6.13 Child's birth order (Phase III)

Nearly half of the children were born prematurely (37/78, 47.4%, three missing information), with gestational ages ranging from 26-38 weeks. Forty-seven children had previously been admitted to hospital prior to PICU admission (Table 6.14).

Number of hospital admissions pre PICU admission	Number of children (%)
None	27 (33.3)
One	18 (22.2)
Two-Four	14 (17.3)
More than five	15 (18.5)
Missing data	7 (8.6)
Total	**81 (99.9)**

Table 6.14 Number of hospital admissions prior to PICU admission (Phase III)

The main reason for hospital admissions was for breathing problems (n=29) and premature birth (n=10). Forty-eight children (59.3%) had been admitted to hospital since PICU discharge (Table 6.15).

Number of hospital admissions post PICU discharge	Number of children (%)
None	29 (35.8)
One	18 (22.2)
Two-Four	20 (24.7)
Five or more	10 (12.3)
Missing data	4 (4.9)
Total	**81 (99.9)**

Table 6.15 Number of hospital admissions post PICU discharge (Phase III)

The main reasons for these hospital admissions, post-PICU discharge, were for respiratory problems (n=24) and non-respiratory illnesses (n=11). Forty children (49.4%) had visited the GP for any reason related to their health in the previous month (Table 6.16); twenty (50%) of these visits were for a respiratory problem.

Number of visits to GP post PICU discharge	Number of children (%)
None	39 (48.1)
One	23 (28.4)
Two-Four	16 (19.8)
Five or more	1 (1.2)
Missing data	2 (2.5)
Total	**81 (100.0)**

Table 6.16 Visits to GP post PICU discharge for any health reason (Phase III)

Parents were asked if they had been told by a health care professional whether their child had a particular health problem (Table 6.17). Responses from at least one parent revealed that 43 parents (35.5%) perceived their child to have a chronic respiratory health problem (Table 6.17). Other health problems described by parents included Coeliac disease, failure to thrive (which could be classified as digestive/gut problems), and a urinary tract infection (which could be classified as a kidney/bladder problem).

Health problem	Number of responses by at least one parent
Asthma	20
Chronic respiratory problem	22
Cystic fibrosis	1
Anxiety problems	1
Behavioural problems	4
Cancer	2
Cerebral palsy	5
Chest abnormality	4
Chronic allergies	3
Developmental delay	26
Digestive/gut problems	22
Down's Syndrome	3
Epilepsy/fits	4
Hearing problems	6
Heart problems	18
Immune problems	8
Kidney/bladder problems	7
Learning problems	19
Liver problems	2
Neurological/brain problems	10
Orthopaedic (bone/joint) problems	5
Sleep problems	10
Speech problems	21
Vision problems	11
Any other health problem	16

Table 6.17 Child's past medical history as perceived by at least one parent (Phase III)

Several children presented with risk factors for respiratory disease, such as previous oxygen therapy (34/81, 42.0%), mechanical ventilation (31/81, 38.3%), and steroid use (21/81, 25.9%). At the time of completing the questionnaire, thirteen children (16.0%) received oxygen therapy at home, day or night (0.1-1.5 litres/minute). Children who had been ventilated previously had been so for 1-105 days (mean=23.7 days, median=11.0 days), 1-24 months previously (mean=16.0 months, median=12.0 months).

6.6.5 Parental demographics

Eighty females and forty-one males responded to the PICQoL Time 1 questionnaires (n=121). Most respondents were the child's biological parent (Table 6.18). Most parents were Caucasian, married and attained GCSE or O levels as their highest educational qualification (Tables 6.19-6.21).

Relationship	Number of respondents (%)
Biological parent	117 (96.7)
Foster parent	2 (1.7)
Grandmother (legal guardian)	1 (0.8)
Missing data	1 (0.8)
Total	**121 (100.0)**

Table 6.18 Relationship to child - all respondents (Phase III)

Ethnic origin	Number of respondents (%)
Caucasian	105 (86.8)
Pakistani	14 (11.6)
Gypsy	1 (0.8)
Missing Data	1 (0.8)
Total	**121 (100.0)**

Table 6.19 Parental ethnic group – all parents (Phase III)

Marital status	Number of respondents (%)
Married	85 (70.3)
Living together	20 (16.5)
Divorced or separated	4 (3.3)
Single	11 (9.1)
Missing Data	1 (0.8)
Total	**121 (100.0)**

Table 6.20 Parental marital status – all parents (Phase III)

Highest educational qualification attained	Number of respondents (%)
No formal education	27 (22.3)
GCSE or o level	48 (39.7)
A level	8 (6.6)
Professional qualification	21 (17.4)
Degree or higher	16 (13.2)
Missing data	1 (0.8)
Total	**121 (100.0)**

Table 6.21 Highest grade of parental educational qualification - all parents (Phase III)

The mean parental age was 31.0 years (SD 7.1 years) with a range of 16-64 years (Figure 6.3); the distribution of age is positively skewed.

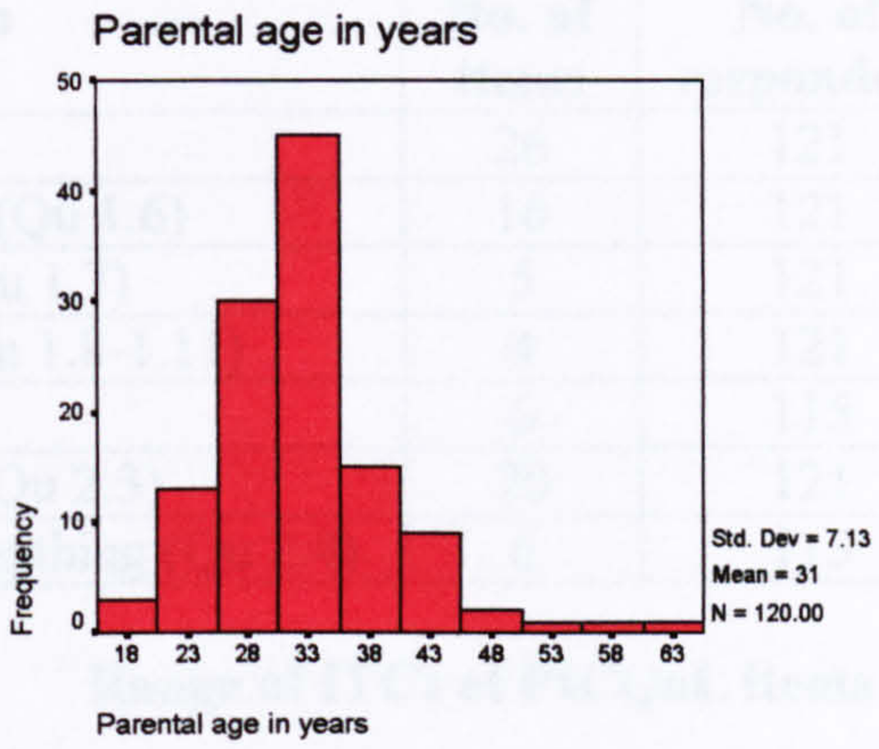


Figure 6.3 Histogram of parental age (years) – all parents (Phase III)

Twenty-two parents said that their work status had changed since their child's PICU admission; ten parents gave up work, and seven parents reduced their working hours because of their child's health problems. Risk factors for respiratory disease were assessed; 31 parents (25.6%) smoked in the home, and 49 parents (40.5%) said they had pets in the home.

6.7 Results 2

6.7.1 Item reduction

Items were removed from the PICQoL questionnaire if the endorsement frequency was outside the range 0.2-0.8, or the ITCs were <0.20 (Streiner & Norman, 1995). Endorsement frequencies for response options of the PICQoL, IPQ and STAI items (Time 1) all fell within an acceptable endorsement range (0.2-0.8), with the exception of sixteen child demographic items. These items included oxygen therapy at home and fifteen past medical history items, with 2-14% of parents not replying 'no'; findings similar to those described earlier (Chapter 5). These items were not removed because of their clinical importance.

6.7.2 Homogeneity of the HRQoL measure

Item-total correlations were calculated for the PICQoL daily activities (Qu. 1.4), IPQ (Qu. 1.6), impact on family (Qu. 1.7) parental worries about health (Qu. 1.8-1.11), STAI (Qu.

1.12), respiratory symptoms (Qu. 2.3) and respiratory limitations (Qu. 2.4) items (Table 6.22).

Question	No. of items	No. of responders	Range of item-total correlations
Daily activities (Qu 1.4)	26	121	0.81-0.85
IPQ minus causal items (Qu 1.6)	16	121	0.50-0.63
Impact on family life (Qu 1.7)	5	121	0.86-0.89
Worries about health (Qu 1.8-1.11)	4	121	0.80-0.86
STAI: Y-6 (Qu 1.12)	6	115	0.84-0.85
Respiratory symptoms (Qu 2.3)	20	121	0.92-0.93
Limitations owing to breathing (Qu 2.4)	6	115	0.79-0.84

Table 6.22 Range of ITCs of PICQoL items (Phase III)

The results revealed that all items correlated with their total scores above 0.2, no items were removed on this basis. Single items (general health, pre and post PICU health comparison, acute illness, general breathing, and comparison of pre and post PICU breathing) were not included in the analysis.

6.7.3 Development of the scoring system

6.7.3.1 General points

Items in the PICQoL questionnaire were recoded to ensure that all items were positively scored so that a higher score represented better health and HRQoL. The number of parents who completed half or more of the items in each scale was determined and scores calculated for these parents. Raw scores were computed by calculating the mean of the items and transforming to a 0-100 scale. The IPQ and STAI scores were calculated using scoring systems published by the scale developers (Marteau & Bekker, 1992; Weinman et al, 1996). Ten scores were developed from the PICQoL items using weights derived from the FA of questions with less than forty items.

6.7.3.2 Single item scores

The child's general health score (Qu. 1.1) and general level of breathing score (Qu. 2.1) were calculated using the Thurstone method of equal-appearing intervals, as utilised in the CHQ (Landgraf et al, 1996). Landgraf et al (1996) report that empirical studies have shown that the interval between 'excellent' and 'very good' is about half the size of the interval

between 'fair' and 'good'. Thus 'excellent' = 5, 'very good' = 4.4, 'good' = 3.4, 'fair' = 2.2 and 'poor' = 1 (Ware et al, 1992). The raw scores were transformed using the formula:

$$\frac{(\text{Raw score} - 1)}{4} \times 100$$

For example: 'Excellent' = (5-1)/4 x 100 = 100; 'very good' = 85; 'good' = 60; 'fair' = 30, and 'poor' = 0.

The child's comparison of health score (Qu. 1.2) and comparison of breathing score (Qu. 2.2) pre and post PICU were calculated in a similar manner to the general health/breathing scores. Responses were recoded so that 'much better now' = 5, 'somewhat better now' = 4, 'the same now' = 3, 'somewhat worse now' = 2 and 'much worse now' =1. However, these scores are equidistant so the Thurstone method was not used. The scores were transformed as described previously so 'much better now' = 100, 'somewhat better now' = 75, 'the same now' = 50, 'somewhat worse now' = 25, and 'much worse now' = 0.

6.7.3.3 Multiple item scores

An impact on family score was calculated for those parents that completed more than half of the items. The responses were coded so that a higher value represented no disruption (or impact) to family life, e.g. 'very often' = 1, 'quite often' = 2, 'occasionally' = 3, 'almost never' = 4, and 'never' = 5. The raw score was calculated using the mean of the items and the raw score was transformed on a 0-100 scale using the formula described previously. Syntax in SPSS 10.0 was used to compute the impact on family score (Appendix XI).

A parental worries about health score was calculated using a similar approach to the impact on family score. Responses were recoded, where 'a lot' = 1, 'quite a bit' = 2, 'some' = 3, 'a little bit' = 4, and 'not at all' = 5. Although the items were described individually in the questionnaire it was logical to group them as a sub-scale on parental worries about health. The raw scores were calculated using the mean and transformed on a 0-100 scale as illustrated previously, using SPSS syntax (Appendix XI).

A respiratory limitations score was calculated by recoding items so that 'not limited at all' = 4, 'sometimes limited' = 3, 'often limited' = 2, and 'always limited' = 1. Scores were calculated for those parents that completed three or more items. Raw scores were calculated and transformed using the formula described previously, but with a denominator

of three. Syntax was developed in SPSS to calculate the respiratory limitations score (Appendix XI).

6.7.4 Factor analysis

6.7.4.1 Daily activity items (Qu. 1.4)

The daily activities question was recoded so that 'much less than other children' = 1, 'somewhat less than other children' = 2, 'about the same as other children' = 3, 'somewhat more than other children' = 4, 'much more than other children' =5, 'not able to do this activity yet' = 3 and 'missing' or 'invalid responses' = median value (calculated as three for every item). Each output was observed for simple structure, namely a factor matrix in which the factors each have a few high loadings (Kline, 1994). Factor analysis was conducted on Phase II (Chapter 5), Phase III and merged Phase II/III data. Prior inspection of the correlation matrices of items from merged data revealed significant correlations in the majority of items suggesting that some items may be related and one or more factors may be formed in a FA (Bryman & Cramer, 2001). Principal components analysis with varimax rotation was performed through SPSS on 26 items from Qu. 1.4 for a sample of 153 parents (merged data from Phase II & III minus duplicate parents) using one-factor, two-factor and three-factor solutions. Orthogonal rotations were used because of their ability to achieve simple structure. The scree plot identifies the cut-off point for factor rotation and is where the line changes slope (Catell, 1952; Kline, 1994). The scree plot from the PCA of merged data suggested two or three possible factors (Figure 6.4).

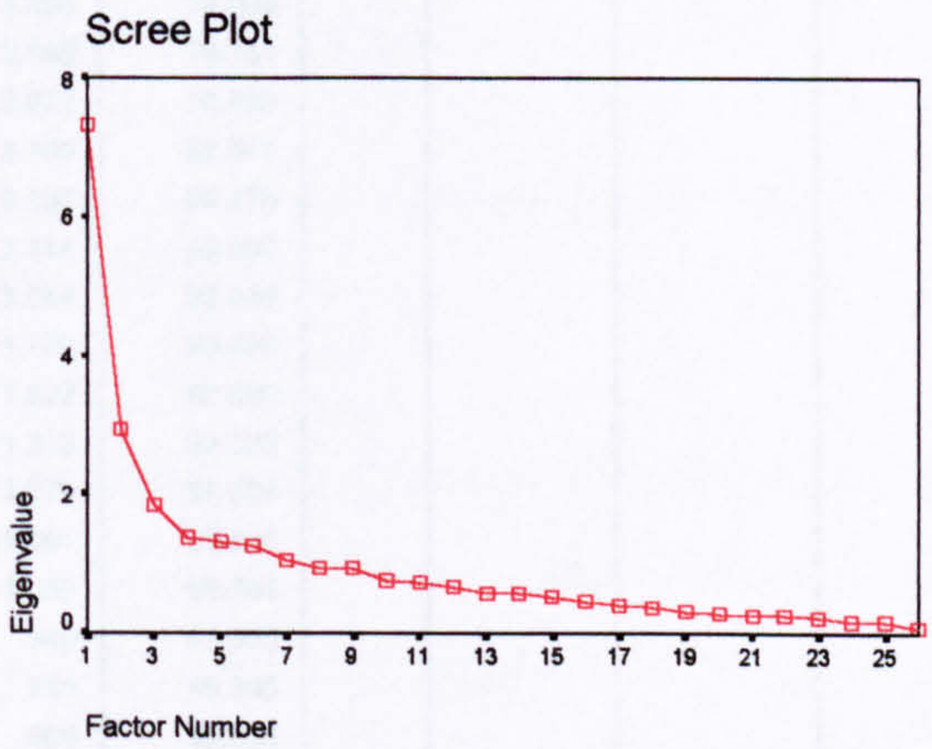


Figure 6.4 Scree plot of PCA daily activity items (Qu. 1.4) – merged Phase II/ III data

In PCA (one-factor solution) with a cut-off factor loading score of 0.3, 73.1% of the items (k = 19) loaded on one factor accounting for 27.1% of the variance (Appendix XII). Some of these items had skewed distributions. A cluster of non-loads related to negative emotional state (quiet, cries, tired, behaves, angry, tantrums and pain) suggesting that there might be extra structure. In PCA three-factor solution (varimax rotation) with a cut-off factor loading score of 0.3, all 26 items loaded on three factors, accounting for 46.1% of the variance, of which 11.1% was due to the third factor (Appendix XII). In this factor a small group of items (excluding the double-loaded items) constituted introverted-type behaviour alongside physical attributes. However, there were too few such items to constitute a robust third factor. Thus, a two-factor solution appeared to be the best choice for PCA.

In PCA with a two-factor solution (varimax) rotation and 0.3 as a cut-off score, the first factor accounted for 27.1% of the variance and the second factor accounted for 11.6% of the variance (Table 6.23).

Total Variance Explained

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	7.045	27.097	27.097	7.045	27.097	27.097	6.858	26.377	26.377
2	3.020	11.614	38.711	3.020	11.614	38.711	3.207	12.334	38.711
3	1.919	7.381	46.093						
4	1.439	5.533	51.626						
5	1.389	5.342	56.968						
6	1.269	4.880	61.848						
7	1.123	4.320	66.168						
8	1.034	3.978	70.146						
9	.898	3.453	73.599						
10	.828	3.185	76.784						
11	.799	3.072	79.856						
12	.647	2.489	82.345						
13	.555	2.133	84.478						
14	.550	2.114	86.592						
15	.534	2.054	88.646						
16	.456	1.752	90.398						
17	.422	1.622	92.020						
18	.341	1.313	93.332						
19	.331	1.272	94.604						
20	.284	1.091	95.695						
21	.262	1.009	96.704						
22	.247	.949	97.653						
23	.185	.711	98.365						
24	.173	.666	99.030						
25	.153	.587	99.617						
26	.100	.383	100.000						

Extraction Method: Principal Component Analysis.

Table 6.23 PCA (2-factor solution) variance of merged data - daily activity items (Qu. 1.4)

Loadings of items on factors were ordered and grouped by size of loading to facilitate interpretation (Table 6.24). The rotated component (varimax) matrix produced five double loads: 'naughty', 'pays attention', 'laughs', 'happy' and 'sad' (Table 6.24).

Rotated Component Matrix[a]

	Component	
	1	2
Holds recoded (medians)	.802	.167
Learns recoded (medians)	.767	-.199
Understands recoded (medians)	.755	-.220
Grasps recoded (medians)	.748	9.765E-02
Plays with friends recoded (medians)	.740	3.847E-02
Moves recoded (medians)	.738	3.065E-02
Speech recoded (medians)	.737	-.271
Walks recoded (medians)	.679	-8.72E-03
Plays with siblings recoded (medians)	.617	.114
Naughty recoded (medians)	.544	.475
Vision recoded (medians)	.495	4.636E-02
Pays attention recoded (medians)	.488	-.325
Laughs recoded (medians)	.441	-.335
Height recoded (medians)	.437	-.276
Plays on own recoded (medians)	.363	-.203
Hearing recoded (medians)	.359	-5.79E-02
Weight recoded (medians)	.351	-.104
Quiet recoded (medians)	-.252	7.995E-02
Tired recoded (medians)	-.201	.176
Cries recoded (medians)	-9.33E-02	.761
Angry recoded (medians)	7.146E-02	.698
Tantrums recoded (medians)	.233	.653
Happy recoded (medians)	.420	-.556
Sad recoded (medians)	-.343	.554
Behaves recoded (medians)	8.429E-02	-.378
Pain recoded (medians)	1.694E-02	.357

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 3 iterations.

Table 6.24 PCA (2-factor solution) rotated factor matrix of merged data - daily activity items (Qu. 1.4)

Items were removed if the difference between the double loads was greater than 0.1. 'Naughty' loaded slightly higher on Factor 1 than Factor 2 (0.544 v. 0.475). Similar loading values for the 'naughty' item may suggest that parents perceived this behaviour in both a positive and negative light; naughty was therefore retained in both factors as it fitted conceptually. 'Pays attention' loaded more highly on Factor 1 than Factor 2 (0.488 v. –0.325) and so was removed from Factor 2. 'Laughs' loaded more highly on Factor 1 than Factor 2 (0.441 v. –0.335) and so was removed from Factor 2. 'Happy' loaded more highly

on Factor 2 than Factor 1 (-0.556 v. 0.420) and so was removed from Factor 1. 'Sad' loaded more highly on Factor 2 than Factor 1 (0.554 v. –0.343) and so was removed from Factor 1. If all double loads had been removed completely from the factors this may have increased the alpha coefficients for the factors by making them more homogenous, however, removing them would have meant reducing the range of behaviour sampled by the factor. 'Tired' and 'quiet' were the only items from Qu. 1.4 that did not load on to a factor, and may warrant exclusion from future questionnaires.

Factor 1 was described as a positive milestones factor (holds, learns, grasps, understands, speech, plays with friends, moves, walks, plays with siblings, naughty, pays attention, vision, laughs, plays on own, hearing, height, weight). Values for items in the component score matrix for each factor were placed in order of size and observed for distinct groupings. Using a weighting scale of a weight as 0.026 in Factor 1, weights were assigned from the component score matrix to reflect the relative value of each component coefficient (Table 6.25), where:

- >0.0125-0.0385 = weight 1
- >0.0385-0.0645 = weight 2
- >0.0645-0.0905 = weight 3
- >0.0905-0.1165 = weight 4
- >0.1165-0.1425 = weight 5

Items were scored positively or negatively depending upon the results of the rotated factor matrix (Table 6.25). The positive milestones score was calculated using the weights from the factor score coefficient matrix:

5 x (holds + grasps) ADD 4 x (moves + walks + speech + learns + understands + plays with siblings + plays with friends + naughty) ADD 3 x (vision) ADD 2 x (height + weight + hearing + pays attention + plays on own + laughs).

The score was transformed on a 0-100 scale using the formula:

$$\frac{\text{Actual score} - \text{minimum score}}{\text{Maximum score} - \text{minimum score}} \times 100$$

The maximum score was calculated as 228 and the minimum score as zero (Appendix XI).

Component Score Coefficient Matrix

	Component	
	1	2
Moves (medians)	.112	.039
Walks (medians)	.102	.024
Grasps (medians)	.117	.061
Holds (medians)	.127	.086
Height (medians)	.055	-.072
Weight (medians)	.049	-.020
Vision (medians)	.077	.035
Hearing (medians)	.052	-.004
Speech (medians)	.100	-.058
Pain (medians)	.017	.116
Sad (medians)	-.030	.165
Happy (medians)	.041	-.162
Angry (medians)	.039	.228
Learns (medians)	.108	-.034
Pays attention (medians)	.061	-.085
Understands (medians)	.105	-.041
Plays on own (medians)	.047	-.051
Plays with siblings (medians)	.098	.061
Plays with friends (medians)	.113	.042
Cries (medians)	.016	.242
Laughs (medians)	.053	-.090
Naughty (medians)	.101	.175
Behaves (medians)	-.002	-.119
Tired (medians)	-.023	.049
Quiet (medians)	-.035	.016
Tantrums (medians)	.061	.220

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

Table 6.25 PCA (2-factor solution) factor score coefficient matrix of merged data - daily activity items (Qu. 1.4)

Factor 2 was described as negative emotions (cries, angry, tantrums, sad, happy, behaves, pain, naughty). Using a weighting scale of 0.06 in Factor 2, weights were assigned using the factor score coefficient matrix (Table 6.25) where:

- >0.220 - 0.280 = weight 4
- >0.160 - 0.220 = weight 3
- >0.100 - 0.160 = weight 2

Items were scored positively or negatively depending upon the results of the rotated factor matrix (Table 6.25). The negative milestones score was calculated using the weights developed from the factor score correlation matrix:

4 x (tantrums + angry + cries) ADD 3 x (sad – happy + naughty) ADD 2 x (pain – behaves)

The score was transformed so that a high score reflected a happy child, thus complying with the scoring system where 100 = best score, 0 = worst score. The negative emotion score was transformed as:

$$\frac{\text{Maximum score} - \text{actual score}}{\text{Maximum score} - \text{minimum score}} \times 100$$

The maximum score was calculated as 80 and the minimum score as –20 (Appendix XI).

6.7.4.2 Respiratory symptom items (Qu. 2.3)

The calculation of the respiratory symptom score was developed from FA of merged data from 153 parents (Phase II/III). The apnoea question was not included in the Phase II questionnaire, so the median response of Phase III parents was coded for missing values of Phase II parents. Syntax in SPSS was developed to recode 'not applicable' in the severity question if the parent had recorded 'not applicable' in the frequency question for each symptom (Appendix XI). However, prior to this analysis, PCA was also performed on Phase II and Phase III data (Appendix XIII) to identify possible factors around which to develop a scoring system.

The results of these earlier factor analyses (2-factor solution) illustrated that frequency and severity of respiratory symptoms were not separate dimensions; there may be sub-dimensions not well reflected by the overall score, e.g. apnoea and snuffles. The PCA of Phase II data showed Factor 1 as a 'miscellaneous symptoms' factor and Factor 2 as a 'cough/snuffles' factor (severity and frequency). The PCA of Phase III data revealed Factor 1 as a 'miscellaneous symptoms' factor and Factor 2 as a 'slow breathing' factor (slow and apnoea). The PCA of merged Phase II/III data revealed Factor 1 as a 'miscellaneous symptom' factor and Factor 2 as a 'cough/snuffles' factor, similar to Phase II factors. All variations of the two-factor PCA therefore showed a 'miscellaneous symptom factor' as the first and much the most important factor. There was no clear and stable second factor in the afore-mentioned analysis that the one-factor solution ignored. Therefore, a single symptom score was developed using the output from the PCA one-factor solution of merged data (Phase II/III) to calculate the weights of individual items.

With a cut-off factor loading score of 0.3, 42.6% of the items loaded on one factor (Table 6.26).

Total Variance Explained

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	8.529	42.646	42.646	8.529	42.646	42.646
2	2.062	10.312	52.959			
3	1.620	8.099	61.057			
4	1.318	6.591	67.648			
5	1.047	5.234	72.882			
6	.998	4.991	77.873			
7	.886	4.430	82.303			
8	.693	3.464	85.767			
9	.666	3.330	89.097			
10	.611	3.056	92.154			
11	.393	1.964	94.117			
12	.278	1.392	95.509			
13	.237	1.186	96.695			
14	.176	.878	97.573			
15	.155	.777	98.350			
16	9.598E-02	.480	98.830			
17	8.412E-02	.421	99.251			
18	6.073E-02	.304	99.555			
19	5.247E-02	.262	99.817			
20	3.663E-02	.183	100.000			

Extraction Method: Principal Component Analysis.

Table 6.26 PCA (1-factor solution) on merged data - respiratory symptom items (Qu. 2.3)

The one-factor solution illustrated that for every symptom, the severity item contributed more to the factor than the frequency item. The snuffles and slow breathing items contributed less than all the other symptoms, except apnoea. The apnoea items contributed little to the combined symptom score, few children had the symptom, and those that did were not necessarily those children with the most severe or frequent other symptoms. Therefore apnoea was removed from the calculation of the respiratory symptom score as the items loaded on the factors <0.3 (Table 6.27). However, advice was sought from expert clinicians who believed that apnoea items must be a significant clinical factor in terms of the child's health and impact on the family and hence the child's HRQoL. The apnoea items were therefore treated as a separate question.

Component Matrix[a]

	Component
	1
Wheeze severity	.847
Stridor severity	.834
Fast breathing severity	.825
Day-time cough severity	.806
Night-time cough severity	.786
Fast breathing frequency	.767
Wheeze frequency	.739
Breathless when active severity	.737
Breathless when still severity	.712
Stridor frequency	.712
Day-time cough frequency	.676
Breathless when active frequency	.643
Night-time cough frequency	.632
Breathless when still frequency	.631
Snuffles frequency	.494
Slow breathing severity	.485
Slow breathing frequency	.427
Snuffles freqeuncy	.301
Apnoea severity	.187
Apnoea frequency	.141

Extraction Method: Principal Component Analysis.

a. 1 components extracted.

Table 6.27 PCA (1-factor solution) component matrix of merged data - respiratory symptom items (Qu. 2.3)

Using a weighting scale of a weight as 0.02 in Factor 1, weights were assigned from the component score matrix (Table 6.28), where:

- > 0.09 = weight 5
- > 0.07 - 0.09 = weight 4
- > 0.05 - 0.07 = weight 3
- > 0.03 - 0.05 = weight 2

Weights were used to reflect the relative value and importance of the coefficients in calculating the respiratory symptom score.

Component Score Coefficient Matrix

	Component
	1
Day-time cough frequency	.079
Day-time cough severity	.095
Night-time cough frequency	.074
Night-time cough severity	.092
Stridor frequency	.083
Stridor severity	.098
Wheeze frequency	.087
Wheeze severity	.099
Fast breathing frequency	.090
Fast breathing severity	.097
Slow breathing frequency	.050
Slow breathing severity	.057
Breathless when still frequency	.074
Breathless when still severity	.084
Breathless when active frequency	.075
Breathless when active severity	.086
Snuffles frequency	.035
Snuffles severity	.058

Extraction Method: Principal Component Analysis.

Table 6.28 PCA (1-factor solution) component score coefficient matrix of merged data - respiratory symptom items (Qu. 2.3)

The respiratory symptom score was calculated as:

5 x (sdtcough + sntcough + sstridor + swheeze + sfaster + sstill + sactive) ADD 4 x (fdtcough + fntcough + fstridor + fwheeze + ffaster + fstill + factive) ADD 3 x (ssnuffles + sslower) ADD 2 x (fsnuffles + fslower)

The maximum respiratory symptom score was calculated as 333 and the minimum score as zero (Appendix XI). The respiratory symptom score was transformed so that a higher score reflected best health and a lower score reflected worst health, using the formula:

$$\text{Respiratory symptom score} = \frac{(\text{Maximum score} - \text{actual score})}{(\text{Maximum score} - \text{minimum score})} \times 100$$

Syntax was developed to calculate this score (Appendix XI).

6.7.4.3 Homogeneity of the factors

Item-total correlations of factors from the FA of daily activities (Qu. 1.4) and respiratory symptoms (Qu. 2.3) were also calculated. The alpha coefficient for the negative emotion factor was low ($\alpha = 0.39$), suggesting that the items in the scale were not all tapping the same underlying area of interest (Jenkinson & McGee, 1998). The alpha coefficients for the milestones factor ($\alpha = 0.89$) and respiratory symptom factor ($\alpha = 0.93$) were satisfactory suggesting that these factors had high internal reliability.

6.8 Results 3

6.8.1 PICQoL score results

The mean PICQoL scores for all parents (n=121), including IPQ and STAI scores, were calculated for Time 1 and Time 2 (Table 6.29). Although the PICQoL scores were not classified into categories, such as 'above average', average' or 'below average', this warrants future consideration to enhance the interpretability of the PICQoL scores by clinicians and parents.

PICQoL scores		
PICQoL Score	**Time 1 – mean (SD)**	**Time 2 – mean (SD)**
General health score	69.3 (25.6)	71.5 (26.6)
Comparison of general health score	79.6 (25.4)	80.1 (23.0)
Positive milestones score	44.9 (12.4)	46.4 (10.5)
Negative emotions score	53.8 (10.7)	53.6 (9.2)
Impact on family score	32.4 (29.5)	19.7 (26.6)
Parental worries about health score	73.4 (27.3)	81.3 (24.8)
General breathing score	63.1 (28.2)	65.1 (32.9)
Comparison of general breathing score	63.1 (28.2)	71.2 (30.7)
Respiratory symptom score	75.1 (22.3)	81.3 (19.0)
Respiratory limitations score	85.1 (20.4)	90.5 (17.9)
IPQ scores		
Symptom score	8.8 (2.9)	8.7 (3.2)
Timeline score	2.5 (0.9)	2.3 (0.7)
Consequences score	3.2 (0.6)	3.1 (0.7)
Cure/control score	2.9 (0.6)	2.9 (0.6)
STAI score		
STAI score	41.8 (15.1)	34.3 (16.6)

Table 6.29 Mean PICQoL scores Time 1 & Time 2 (Phase III)

6.8.2 Reliability

6.8.2.1 Internal consistency

The internal consistency was calculated using Cronbach's alpha statistic, with alpha values falling within an acceptable range (0.7-0.9) with the exception of respiratory symptoms (α=0.93) (Table 6.30):

Question	Number of responders	Cronbach's alpha statistic
Activities (Qu1.4)	121	0.83
IPQ minus causal items (Qu. 1.6)	121	0.57
Impact on family life (Qu 1.7)	121	0.90
Worries about health (Qu1.8-1.11)	121	0.87
STAI: Y-6 (Qu. 1.12)	115	0.86
Respiratory symptoms (Qu 2.3)	121	0.93
Limitations owing to breathing (Qu 2.4)	115	0.84

Table 6.30 Internal consistency of PICQoL items (Phase III)

6.8.2.2 Test-retest reliability

The test-retest reliability of the PICQoL and HUI questionnaires was calculated via Pearson (and Spearman's rho) correlations (Table 6.31). Time 1 and Time 2 PICQoL scores revealed moderate to high correlations (range 0.510 - 0.773). McDowell and Newell (1996) state that a figure of 0.7 represents a high correlation, 0.6 a medium correlation and less than 0.5 a low correlation.

Score	Pearson correlation (2-tailed)	Spearman's rho correlation
General health score (Qu. 1.1)	0.634**	0.629**
Comparison of general health score (Qu. 1.2)	0.588**	0.584**
Milestones score (Qu. 1.4)	0.862**	0.698**
Negative emotions score (Qu. 1.4)	0.781**	0.771**
IPQ symptom score (Qu. 1.5)	0.662**	0.714**
IPQ timeline score (Qu. 1.6)	0.690**	0.694**
IPQ consequences score (Qu. 1.6)	0.682**	0.683**
IPQ cure/control score (Qu. 1.6)	0.577**	0.643**
Impact on family score (Qu. 1.7)	0.786**	0.773**
Worries about health score (Qu. 1.8-1.11)	0.822**	0.776**
STAI score (Qu. 1.12)	0.624**	0.648**
General breathing score (Qu. 2.1)	0.725**	0.728**
Comparison of general breathing score (Qu. 2.2)	0.529**	0.510**
Respiratory symptom score (Qu. 2.3)	0.672**	0.719**
Respiratory limitations score (Qu. 2.4)	0.756**	0.791**

** Correlation is significant at the 0.01 level (2-tailed)

Table 6.31 Correlations for PICQoL scores Time 1 & Time 2 (Phase III)

The correlations were repeated for the HUI variables Time 1 and Time 2 (Table 6.32). Responses to Time 1 and Time 2 HUI questions revealed low to high correlations (range 0.398 - 0.846). Slight differences were observed between Pearson and Spearman's rho correlations for both PICQoL and HUI correlations. Most of the PICQoL scores were non-normally distributed; the non-parametric correlation (Spearman's rho) was preferred (Streiner & Norman, 1995).

Question	Pearson correlation (2-tailed)	Spearman's rho correlation
Qu. 1 vision	0.805**	0.805**
Qu. 2 vision	0.581**	0.581**
Qu. 3 hearing	0.410**	0.398**
Qu. 4 hearing	0.594**	0.596**
Qu. 5 speech	0.730**	0.732**
Qu. 6 speech	0.784**	0.765**
Qu. 7 emotion	0.859**	0.846**
Qu. 14 emotion	0.583**	0.578**
Qu. 8 pain	0.773**	0.471**
Qu. 15 pain	0.713**	0.452**
Qu. 9 ambulation (mobility)	0.843**	0.841**
Qu. 10 dexterity (mobility)	0.408**	0.434**
Qu. 11 cognition	0.747**	0.733**
Qu. 12 cognition	0.481**	0.373**
Qu. 13 self-care	0.837**	0.802**
Qu. 16 general health	0.567**	0.561**
Qu. 19 time to complete questionnaire	0.596**	0.506**

** Correlation is significant at the 0.01 level (2-tailed)

Table 6.32 Correlations for HUI questions Time 1 & Time 2 (Phase III)

6.8.2.3 Reliability of the IPQ

The results and interpretation of the FA (PAF) of sixteen IPQ items for merged Phase II/III data are presented (Appendix XIV). Merged data was utilised to increase the sample size for the analysis. The scree plot from the PAF of merged data suggested that there might be two or three factors (Figure 6.5).

'Original in colour'

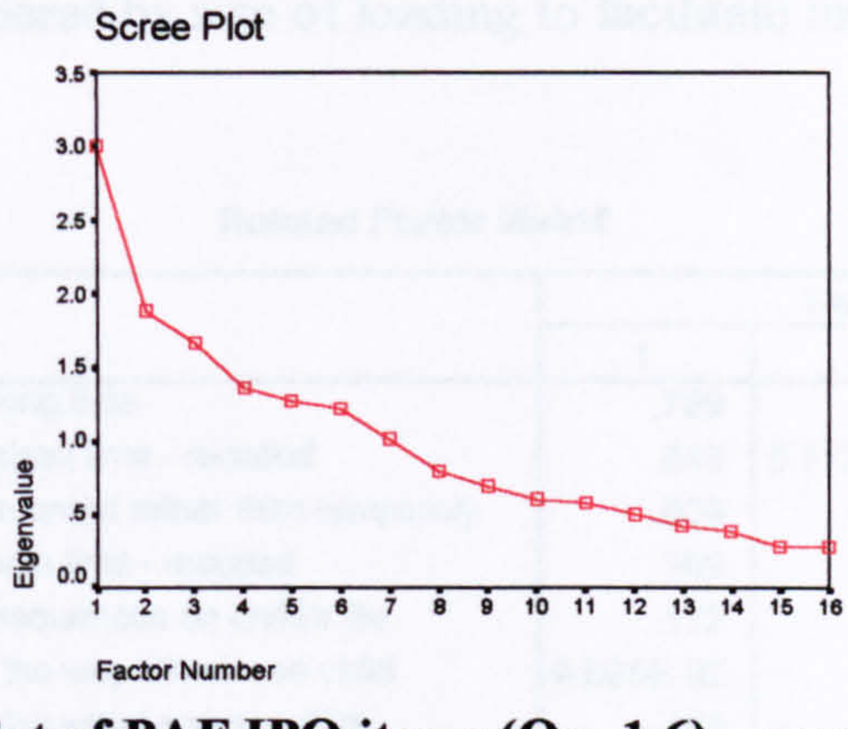


Figure 6.5 Scree plot of PAF IPQ items (Qu. 1.6) – merged Phase II/III data

In the PAF, one-factor solution, with a cut-off factor loading score of 0.3, 50.0% (k=8) of the items loaded on one factor accounting for 22.5% of the variance (Appendix XIV). A cluster of non-loads related to consequences and cure/control, suggesting that there might be extra structure. In PAF, two-factor solution (varimax rotation) with a cut-off factor loading score of 0.3, fourteen items loaded on two factors, accounting for 30.6% of the variance (Appendix XIV). Factor 1 described a miscellaneous consequences and cure/control factor, and Factor 2 described a timeline factor, the latter factor replicating the original IPQ timeline factor. In the PAF, three-factor solution (varimax rotation) and 0.3 cut-off factor loading score, the first factor accounted for 18.8% of the variance, the second factor as 11.8% of the variance, and the third factor as 10.5% of the variance (Table 6.33).

Total Variance Explained

	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
Factor	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	3.013	18.834	18.834	2.411	15.071	15.071	1.825	11.408	11.408
2	1.889	11.805	30.638	1.195	7.467	22.537	1.740	10.876	22.284
3	1.672	10.452	41.090	1.081	6.755	29.293	1.121	7.009	29.293
4	1.361	8.503	49.593						
5	1.274	7.961	57.554						
6	1.220	7.625	65.178						
7	1.020	6.375	71.554						
8	.795	4.971	76.525						
9	.699	4.368	80.892						
10	.604	3.772	84.664						
11	.573	3.582	88.247						
12	.497	3.106	91.353						
13	.430	2.685	94.038						
14	.385	2.408	96.446						
15	.289	1.805	98.252						
16	.280	1.748	100.000						

Extraction Method: Principal Axis Factoring.

Table 6.33 PAF (3-factor solution) variance of merged data of IPQ items (k=16)

Loadings of items were ordered by size of loading to facilitate interpretation; there were no double loads (Table 6.34).

Rotated Factor Matrix[a]

	Factor		
	1	2	3
Illness would last a long time	.769	.232	-.142
Illness would last a short time - recoded	.645	6.713E-02	-6.87E-02
Illness would be permanent rather than temporary	.609	.149	3.601E-02
Illness will improve with time - recoded	.369	-.164	.103
Illness has had consequences on child's life	.172	.614	.176
Illness has affected the way others see child	9.895E-02	.514	-.150
Illness has affected the way I see my child	-.145	.482	-9.93E-02
Illness has had economic consequences	.232	.461	-8.25E-02
Child could control his illness	-5.55E-03	.419	-.175
Illness has had little effect on child's life - recoded	.287	.409	.283
Illness was serious	8.150E-02	.309	6.796E-02
Child could control symptoms	-8.46E-02	.176	2.567E-02
Little could be done to improve illness - recoded	-6.35E-02	-8.47E-02	.790
Illness has become easier to live with - recoded	.153	-4.61E-03	.336
Child's recovery due to chance - recoded	-.212	-.228	.334
Treatment would be effective	-.165	.133	.223

Extraction Method: Principal Axis Factoring.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 5 iterations.

Table 6.34 PAF (3-factor solution) rotated factor matrix of merged IPQ items (k=16)

Factor 1 replicated the IPQ timeline factor, and Factor 2 described a consequences factor. Factor 2 replicated the IPQ consequences factor with the exception of one item 'child could control his illness', which is a cure/control item in the IPQ. Factor 3 described a cure/control factor, which replicated the IPQ cure/control factor with the exception of one item, 'illness has become easier to live with', which is a consequences item in the IPQ. The FA therefore reproduced similar factors to those described by the IPQ developers, thus contributing to its reliability in this population.

6.9 Validity

6.9.1 HUI results

One hundred and fourteen parents responded to the HUI Time 1 questionnaire, representing 40 male and 74 female parents. The mean child age on parental completion of the HUI questionnaires was 22.3 months (range 1-59 months). The majority of parents (Time 1) scored the first 'best function' or last 'not applicable' response options for each HUI item (Table 6.35).

HUI item (attribute)	Number of parents recording first response option (%)	Number of parents recording last response option (%)
Qu. 1 vision (sensation)	29 (26)	80 (70)
Qu. 2 vision (sensation)	59 (52)	50 (44)
Qu. 3 hearing (sensation)	51 (45)	60 (53)
Qu. 4 hearing (sensation)	66 (58)	44 (39)
Qu. 5 speech (sensation)	22 (20)	56 (50)
Qu. 6 speech (sensation)	26 (23)	56 (50)
Qu. 7 emotion (emotion)	72 (63)	16 (14)
Qu. 8 pain (pain)	70 (61)	14 (12)
Qu. 9 ambulation (mobility)	45 (40)	47 (42)
Qu. 10 dexterity (mobility)	78 (70)	18 (16)
Qu. 11 cognition (cognition)	27 (24)	78 (69)
Qu. 12 cognition (cognition)	12 (11)	93 (82)
Qu. 13 self-care (self-care)	21 (19)	71 (63)
Qu. 14 emotion (emotion)	73 (65)	25 (22)
Qu. 15 pain (pain)	73 (65)	11 (10)

Table 6.35 Number of parents responding to first and last response options for HUI items (Time1) Phase III

The mean completion time for HUI (Time 1) (n=111) was 8.4 minutes (range 2-60 minutes) and 7.0 minutes (range 2-35 minutes) for Time 2 (n=53). Utility scores were calculated from eligible responses in Time 1 and Time 2, where a score of one represents 'perfect health' and a score of zero represents 'death' (Table 6.36). Utility scores could only be calculated for six parents in Time 1 (child's age range 37-59 months) and five parents (child's age range 26-59 months) in Time 2. This might suggest that parents of younger children (< 2 years) had difficulty interpreting and completing the HUI items.

Child ID	HUI II (Time 1)	HUI III (Time 1)	Child's age (months)	Child ID	HUI II (Time 2)	HUI III (Time 2)	Child's age (months)
65	0.78	0.53	37	12	0.97	0.86	42
49	0.79	0.67	43	26	1.0	1.0	26
64	0.85	0.74	49	43	1.0	1.0	59
74	0.97	1.0	56	43	1.0	1.0	59
43	1.0	1.0	59	74	1.0	1.0	56
43	1.0	1.0	59				

Table 6.36 HUI II and III utility scores (Time 1/Time 2) with child's age (Phase III)

6.9.2 Criterion validity

The PICQoL and HUI questionnaires have similar items on general health; these scores were correlated (Table 6.37).

PICQoL score & HUI question/attribute (number of respondents)	Pearson correlation	Spearman's rho correlation
PICQoL general health score & HUI general health question (108) [Time 1]	-0.630**	-0.624**
PICQoL general health score & HUI general health question (53) [Time 2]	-0.776**	-0.809**

** Correlation is significant at 0.01 level (2-tailed)

Table 6.37 General health score correlations PICQoL and HUI questionnaires (Phase III)

The PICQoL milestones and negative emotion scores were also correlated with similar HUI attributes for Time 1 and Time 2 (Tables 6.38-6.39).

PICQoL score & HUI attribute (Number of respondents)	Pearson correlation	Spearman's rho correlation
PICQoL milestone score & HUI II sensation attribute (20)	-0.214	-0.163
PICQoL milestone score & HUI II mobility attribute (58)	-0.823**	-0.711**
PICQoL milestone score & HUI II cognition attribute (14)	-0.797**	-0.539*
PICQoL milestone score & HUI II self-care attribute (38)	-0.672**	-0.669**
PICQoL milestone score & HUI III vision attribute (28)	-0.338	-0.148
PICQoL milestone score & HUI III hearing attribute (49)	-0.060	-0.123
PICQoL milestone score & HUI III speech attribute (50)	-0.516**	-0.478**
PICQoL milestone score & HUI III ambulation attribute (63)	-0.783**	-0.700**
PICQoL milestone score & HUI III dexterity attribute (92)	-0.649**	-0.587**
PICQoL milestone & HUI III cognition attribute (14)	-0.792**	-0.539*
PICQoL negative emotion score & HUI II emotion attribute (85)	-0.414**	-0.324**
PICQoL negative emotion score & HUI II pain attribute (98)	-0.131	-0.124
PICQoL negative emotion score & HUI III emotion attribute (96)	-0.075	0.165
PICQoL negative emotion score & HUI II pain attribute (96)	-0.139	-0.206*

* Correlation is significant at the 0.05 level (2-tailed)

** Correlation is significant at the 0.01 level (2-tailed)

Table 6.38 Correlation statistics of PICQoL scores and HUI attributes (Time 1) Phase III

PICQoL score & HUI attribute (Number of respondents)	Pearson correlation	Spearman's rho correlation
PICQoL milestone score & HUI II sensation attribute (11)	-0.777	-0.101
PICQoL milestone score & HUI II mobility attribute (32)	-0.840**	-0.527**
PICQoL milestone score & HUI II cognition attribute (8)	a	.
PICQoL milestone score & HUI II self-care attribute (19)	-0.571*	-0.509*
PICQoL milestone score & HUI III vision attribute (12)	a	.
PICQoL milestone score & HUI III hearing attribute (25)	a	.
PICQoL milestone score & HUI III speech attribute (25)	-0.106	-0.147
PICQoL milestone score & HUI III ambulation attribute (33)	-0.897**	-0.589**
PICQoL milestone score & HUI III dexterity attribute (46)	-0.717**	-0.351*
PICQoL milestone & HUI III cognition attribute (8)	a	.
PICQoL negative emotions score & HUI II emotion attribute (44)	-0.314*	-0.218
PICQoL negative emotions score & HUI II pain attribute (48)	-0.169	-0.121
PICQoL negative emotions score & HUI III emotion attribute (43)	-0.113	-0.006
PICQoL negative emotions score & HUI III pain attribute (55)	-0.204	-0.232

* Correlation is significant at the 0.05 level (2-tailed)

** Correlation is significant at the 0.01 level (2-tailed)

a Cannot be computed as only one attribute level defined and too few observations recorded

Table 6.39 Correlation statistics of PICQoL scores and HUI attributes (Time 2) Phase III

Results revealed moderate correlations in both Time 1 and Time 2, with the exception of PICQoL milestone score and HUI II sensation/HUI III vision and hearing attributes (Time 1); PICQoL negative emotion scores and pain (HUI II/III) and emotion (HUI III) attributes (Time 1 and Time 2); and PICQoL milestone score and HUI III speech attribute (Time 2); all of which were poorly correlated. Some correlations failed to compute in Time 2; this might be owing to only one level of attribute defined and varying sample sizes. There may also have been many missing values for one of the variables and the remaining valid cases all had the same score.

6.9.3 Construct validity

This was assessed by testing the afore-mentioned hypotheses (see 6.1.1). The hypotheses, which specified relationships between the child's illness type and levels of health, levels of breathing, milestones, child emotions, parental anxiety, impact on family life and daily activities were explored statistically by calculating mean PICQoL scores (Table 6.40). Further results concerning hypotheses relating to parental anxiety and illness perceptions are presented (6.9.3.1 and 6.9.3.2).

Type of PICQoL score	Mean scores by illness type (SD)		
	Acute respiratory	Acute & chronic respiratory	Acute respiratory & other chronic illness
General health	76.3 (24.8)	68.6 (26.7)	57.8 (22.2)
Comparison of general health	76.6 (26.3)	84.7 (21.3)	82.4 (25.6)
Positive milestones	49.4 (9.1)	42.1 (13.9)	38.7 (13.5)
Negative emotions	54.6 (10.8	52.4 (8.2)	53.3 (11.8)
Impact on family	20.2 (22.8)	38.0 (32.9)	50.5 (28.3)
Parental worries	78.8 (26.8)	71.3 (26.6)	64.9 (26.9)
General breathing	66.8 (30.4)	56.9 (25.7)	59.6 (24.6)
Comparison of general breathing	71.5 (25.9)	86.1 (17.6)	80.6 (24.0)
Respiratory symptom	80.1 (21.8)	67.6 (22.1)	70.7 (21.7)
Respiratory limitations	92.2 (14.7)	77.5 (22.2)	77.0 (23.5)

Table 6.40 Mean PICQoL scores by illness type (Time 1) Phase III

Parents of acutely ill children perceived their child's general health and general breathing to be better than did parents of children who were acutely ill and had other chronic illnesses. Parents of acutely ill children described less respiratory symptoms and less impact of respiratory symptoms on their child's daily life than parents of children who were chronically ill, as illustrated by the higher respiratory symptom and respiratory limitation scores. However, the impact on family scores were lower for parents of acutely ill children suggesting that for these parents their child's health impacted upon family life more greatly than it did for children with a chronic illness. Parents of acutely ill children had higher positive milestones and negative emotion scores than parents of children with a chronic illness. This suggests that acutely ill children had more 'normal' developmental milestones and were less distressed than chronically ill children. All hypotheses were confirmed in the expected direction, with the exception of impact on family scores and child's illness type.

6.9.3.1 Anxiety

The STAI: Y-6 has been previously validated and the scoring system reported elsewhere (Marteau & Bekker, 1992). In this study, the STAI scale was used to assess a parent's level of anxiety about their child's current health and wellbeing. The STAI scores were calculated for those parents who completed three or more of the STAI items. Comparison of mean STAI scores by parental sex revealed that mothers had a higher mean score (43.1,

SD 15.7) than fathers (39.5, SD 13.9). A paired samples t-test of STAI scores Time 1 and Time 2 (n=51) revealed a significant difference in mean scores (Table 6.41).

Mean scores (SD)	Time 1	Time 2	t-test	
			95% CI for difference	Sig.
STAI score	39.5 (15.4)	34.1 (16.0)	(-9.2, -1.6)	0.006

Table 6.41 Mean (SD) STAI scores and t-test by Time 1/Time 2 responders (Phase III)

Parents of acutely ill children had lower mean STAI scores than parents of chronically ill children and lower parental worries about health scores (Table 6.42). The hypothesis that parents of children with an acute respiratory illness are less anxious about their child's health than parents of children with an acute and chronic respiratory illness was confirmed.

Type of PICQoL score	Mean scores by illness type (SD)		
	Acute respiratory	Acute & chronic respiratory	Acute respiratory & other chronic illness
Worries about health	78.8 (26.8)	71.3 (26.6)	64.9 (26.9)
STAI: Y-6 score	35.8 (15.2)	48.2 (13.1)	49.1 (11.1)

Table 6.42 Mean (SD) STAI and parental worry scores by illness type (Time 1) Phase III

6.9.3.2 Illness perceptions

A symptom score (range 0-13), timeline, consequences and cure control scores (range 0-5) were calculated. The median symptom score for all parents was 9.0 (range 1-13) for PICQoL Time 1; this did not differ by parental sex. The median symptom score for PICQoL Time 2 parents was 10.0 (range 2-13), this was the same for fathers, but the median score for mothers was 8.5. The mean timeline, consequences and cure/control scores were calculated for Time 1 and Time 2 (Tables 6.43-6.44).

IPQ score (Time 1)	All parents Mean (SD)	Male Mean (SD)	Female Mean (SD)
Timeline	2.54 (0.87)	2.25 (0.83)	2.69 (0.86)
Consequences	3.20 (0.65)	3.20 (0.66)	3.20 (0.64)
Cure/control	2.86 (0.60)	3.02 (0.50)	2.78 (0.63)

Table 6.43 IPQ mean (SD) scores by parental sex (Time 1) Phase III

IPQ score (Time 2)	All parents Mean (SD)	Male Mean (SD)	Female Mean (SD)
Timeline	2.28 (0.69)	2.15 (0.58)	2.34 (0.74)
Consequences	3.10 (0.69)	2.98 (0.65)	3.16 (0.71)
Cure/control	2.90 (0.63)	3.13 (0.56)	2.78 (0.64)

Table 6.44 IPQ mean (SD) scores by parental sex (Time 2) Phase III

An independent samples t-test of Time 1 IPQ scores by parental sex revealed a significant difference in mean timeline scores (P=0.008) and cure/control scores (P=0.033) (Table 6.45); no significant differences were found in mean scores between mothers and fathers in Time 2.

IPQ scores	Mothers (n=79/80) Mean (SD)	Fathers (n=41) Mean (SD)	t-test	
			95% CI for difference	Sig.
IPQ illness identity (symptom score)	8.9 (2.9)	8.6 (2.8)	(-1.4, +0.8)	0.571
IPQ timeline	2.7 (0.9)	2.3 (0.8)	(-0.7, -0.1)	0.008
IPQ consequences	3.2 (0.6)	3.2 (0.7)	(-0.2, +0.2)	0.994
IPQ cure/control	2.8 (0.6)	3.0 (0.5)	(0.0, +0.5)	0.033

Table 6.45 Mean (SD) IPQ scores and t-test by parental sex (Time 1) Phase III

A paired samples t-test of IPQ scores Time 1 and Time 2 revealed a significant difference in mean scores for consequences scores only (P=0.01) (Table 6.46).

IPQ scores T1 & T2	Mean (SD)	t-test	
		95% CI for difference	Sig.
IPQ illness identity (symptom score) (n=55)	0.1 (2.5)	(-0.6, +0.8)	0.747
IPQ timeline (n=54)	-0.0 (0.6)	(-0.2, +0.1)	0.533
IPQ consequences (n=55)	-0.2 (0.5)	(-0.3, -0.0)	0.011
IPQ cure/control (n=55)	0.1 (0.6)	(-0.0, +0.3)	0.134

Table 6.46 Paired samples t-test of Time 1 and Time 2 IPQ scores (Phase III)

The relationship between IPQ scores and child's illness type was further explored statistically by calculating means and one-way ANOVA statistics to assess group

differences in illness cognitions (Tables 6.47-6.48). Results revealed significant differences in timeline (f=8.34, df=2,119, P<0.001) and consequences (f=5.83, df=2,120, P=0.004) scores in Time 1 and cure/control scores (f=3.61, df=2,52, P=0.03) in Time 2.

IPQ score	Acute respiratory		Acute respiratory & chronic respiratory		Acute respiratory & other chronic illness		F value	Sig.
	X	95% CI	X	95% CI	X	95% CI		
Symptom	8.6	7.9-9.3	8.8	7.1-10.6	9.0	8.1-9.9	0.2	0.747
Timeline	2.3	2.1-2.57	2.6	2.2-3.0	3.0	2.6-3.3	8.3	<0.001
Consequences	3.0	2.9-3.2	3.5	3.2-3.8	3.4	3.2-3.6	5.8	0.004
Cure/control	2.9	2.8-3.1	3.0	2.7-3.2	2.7	2.4-3.0	2.8	0.068

Note: X = mean; CI = confidence interval

Table 6.47 ANOVA of IPQ scores and child's illness type (Time 1) Phase III

IPQ score	Acute respiratory		Acute respiratory & chronic respiratory		Acute respiratory & other chronic illness		F value	Sig.
	X	95% CI	X	95% CI	X	95% CI		
Symptom	8.2	7.1-9.3	6.2	2.0-10.4	10.5	9.5-11.6	6.1	0.004
Timeline	2.1	1.9-2.4	2.2	2.8-3.2	2.6	2.2-3.0	2.6	0.086
Consequences	3.0	2.8-3.2	3.2	2.4-4.0	3.3	2.9-3.7	1.1	0.362
Cure/control	3.0	2.8-3.2	3.2	2.8-3.6	2.6	2.3-2.9	3.6	0.034

Note: X = mean; CI = confidence interval

Table 6.48 ANOVA of IPQ scores and child's illness type (Time 2) Phase III

Parents of children (Time 1) with an acute respiratory illness perceived their child's illness to last for a shorter time than did parents of children with a chronic respiratory illness, as reflected by the respective timeline scores (2.3 v. 2.6; 2.3 v. 3.0). Parents of children (Time 1) with an acute respiratory illness perceived their child's illness to be less controllable and curable than parents of children with an acute and chronic respiratory illness. However, parents of children with an acute respiratory and other chronic illness perceived their child's illness to be less curable and controllable than parents of children with an acute respiratory illness. Parents of children with an acute and chronic respiratory illness or acute respiratory and other chronic illness perceived their child's illness to have more consequences on their child's life and family life than did parents of children with an acute respiratory illness. The hypothesis that differences in illness perceptions would be seen according to child' illness type were confirmed in the expected direction, with the exception of cure/control scores.

6.9.4 Convergent validity

This was assessed by observing the correlation of the parental worries about health score with another measure of a similar construct, anxiety, using the STAI score (Table 6.49):

PICQoL score (Number of respondents)	Pearson correlation (2-tailed)	Spearman's rho correlation
Parental worry and STAI Time 1 (118)	-0.592**	-0.615**
Parental worry and STAI Time 2 (53)	-0.569**	-0.715**

** Correlation is significant at the 0.01 level (2-tailed).

Table 6.49 Correlation of parental worry and STAI scores (Time 1/Time 2) Phase III

Results revealed that the parental worries score correlated with the construct anxiety as measured by the STAI, establishing convergent validity of the PICQoL questionnaire.

6.10 Practicality

Most parents (n=49) rated the PICQoL questionnaire as 'quite easy' to complete (Table 6.50). The completion times for both questionnaires were wide ranging. The mean completion time for the PICQoL questionnaires for Time 1 was 22.4 minutes (range 4-60 minutes, median = 20.0 minutes). Parents took 2-60 minutes to complete the HUI questionnaire in Time 1 (mean = 8.4 minutes, median = 5.0 minutes).

Ease of completion of PICQoL questionnaire	Number (%)
Very easy	36 (29.8)
Quite easy	49 (40.5)
Neither easy nor difficult	27 (22.3)
Quite difficult	6 (5.0)
Difficult	2 (1.6)
Missing data	1 (0.8)
Total	**121 (100.0)**

Table 6.50 PICQoL questionnaire evaluation (Time 1) Phase III

6.11 Discussion

This survey investigated the reliability (internal, homogeneity, and test-retest), validity (criterion, construct and convergent), practicality and feasibility of the PICQoL

questionnaire in a population of parents whose children were admitted to PICU with a respiratory illness.

6.11.1 Item selection

Rigorous procedures were adhered to in item selection for the HRQoL measure by calculating endorsement rates, ITCs and FA, as in the previous phase. Endorsement rates were within the acceptable range (0.2-0.8) and no items were removed from the PICQoL questionnaire. Item total correlations of the daily activity (Qu. 1.4), IPQ minus causal (Qu. 1.6), impact on family (Qu. 1.7), worries about health (Qu. 1.8-1.11), STAI (Qu. 1.12), respiratory symptoms (Qu. 2.3) and respiratory limitations (Qu. 2.4) items also revealed correlations within an acceptable range (0.50-0.93), these items were retained in the PICQoL questionnaire. The FA of daily activity items (Qu. 1.4) suggested that two items be removed in future versions of the questionnaire.

6.11.2 Reliability

The internal consistency of the HRQoL measure was high (α = 0.83-0.87) for the daily activity, impact on family life, worries about health, STAI, and respiratory limitations items. Alpha coefficients for these items were within an acceptable range, i.e. above 0.7, and not higher than 0.9 (Nunnally, 1978; Streiner & Norman, 1995). The internal consistency of the IPQ items (minus causal items) was low (α = 0.57) and was high (α = 0.93) for the respiratory symptoms items. These results should be interpreted with caution, as alpha coefficients are dependent not only on the magnitude of the correlations among items, but also on the number of items in the scale. A scale can be made to look more 'homogeneous' simply by doubling the number of items, even though the average correlation remains the same (Streiner & Norman, 1995). If alpha coefficients are too high, it may suggest a high level of item redundancy, this may indicate that some of the items are unnecessary, and that the scale as a whole may be too narrow in its scope to have much content validity (Streiner & Norman, 1995). The homogeneity of the measure was satisfactory with ITCs reported within an acceptable range.

The test-retest reliability of the HRQoL measure was good with Time 1 and Time 2 PICQoL scores revealing moderate to good correlations (Pearson correlation 0.529-0.822; Spearman's rho 0.510-0.791) significant at the 0.01 level (2-tailed). Large correlations

indicate reliability, whereas a small correlation may indicate either unreliability or some children getting better and some getting worse, or both. A test-retest reliability of ≥0.8 is generally considered quite sound, although the interpretation of this figure depends on the context in which the measure is to be applied (Polgar & Thomas, 1998).

The Pearson correlation coefficient is preferred over the intraclass correlation coefficient (ICC) in this context as there are two measurements per respondent where Time 1 is more recent to the PICU admission than Time 2, rather than two different observers. The Pearson correlation is a better measure than the ICC in this context because any systematic difference between Time 1 and Time 2 measurements is more likely to be as a result of shifts in the true underlying measurement rather than reliability of the measure. For example, parental anxiety decreasing because the PICU admission is less recent and the children were getting better. When time is used as a fixed factor in intraclass correlation, this will cope with a uniform drop in the scores between Time 1 and Time 2, but only part of the variation will be removed if the drop is a constant percentage. Calculating the Pearson correlation addresses how well the observed score represents a 'real' underlying score for the child at a given point in time. However, ICC's are useful when there are three observers, e.g. parent, child and clinician perspective on HRQoL (Streiner & Norman, 1995). Many of the PICQoL scores were non-normally distributed, so the Spearman's rho correlation coefficient was the correlation of choice.

6.11.3 Validity

Although no 'gold standard' for HRQoL exists, instances occur in which a specific target for an HRQoL measure exists that can be treated as a criterion or 'gold standard'. In these circumstances, one determines whether a measure is measuring what is intended using criterion validity (Guyatt et al, 1993). The HUI was used as the 'gold standard' in this case. When no 'gold standard' exists, other validation strategies can be used, such as content and construct validity (Guyatt et al, 1993).

In the assessment of criterion validity, results revealed a significant (0.01 level, 2-tailed) moderate correlation with the PICQoL general health score and HUI general health item for Time 1 and Time 2 (Pearson correlation = -0.630 & -0.776 respectively). Similar significant correlations were found between the Time 1 PICQoL milestones and negative

emotions scores and HUI II/III attributes (mobility, cognition, self-care, speech, ambulation, dexterity, and emotion). This suggests that the criterion validity of the PICQoL questionnaire was established and favourable against the best available 'gold standard' measure, the HUI.

In the assessment of construct validity all hypotheses were confirmed. However, two hypotheses were not confirmed in the expected direction. Differences in parental illness perceptions were expected according to the child's illness type, where parents of a child with an acute illness would perceive their child's illness to be more controllable and curable than parents of a child with a chronic illness. The reverse was found, parents of children with an acute respiratory and other chronic illness perceived their child's illness to be less curable and controllable than parents of children with an acute respiratory illness. This may be a reflection of the poor prognosis for those children who had other chronic illnesses compared to those children with a chronic respiratory illness whose outcome may have been more favourable. Differences in parental perceptions of the child's illness on the impact of family life were expected according to the child's illness type, where children with an acute and chronic respiratory illness will impact upon family life more so than children with an acute respiratory illness. The reverse was found, the impact on family scores were lower for parents of acutely ill children suggesting that the child's health impacted upon family life more so than for those children with a chronic respiratory illness. This finding may reflect the differences in parental coping styles between parents of acutely ill children and chronically ill children. It may also reflect that the 'impact' on family life might compare differences from the usual, so that the effect of chronic illness is counted as 'usual'. The parents of chronically ill children might also be more used to the health care system. Although the results show promising construct validity of the HRQoL measure, the examination of other constructs is recommended. The more frequently a measure is used, and the more situations in which it performs as expected, the greater the confidence will be in its validity (Guyatt et al, 1993).

The correlation of the PICQoL parental worries about health and STAI scores revealed significant (0.01 level, 2-tailed) correlations (Pearson correlation –0.569 to -0.592, Time 1), demonstrating convergent validity. This finding demonstrates that the parental worries about health items are reliable in assessing parental anxiety, and it might therefore be appropriate to remove the STAI items from future PICQoL questionnaires.

6.11.4 Practicality

The majority of parents (n=85) indicated that the PICQoL questionnaire was 'quite easy' or 'very easy' to complete. The mean completion time (Time 1) was fair at 22.4 minutes. However, it must be acknowledged that the PICQoL questionnaire measure incorporated two other measures, namely the IPQ (39 items) and the short-form STAI (6 items). These measures were incorporated into the HRQoL measure to explore parental illness perceptions and parental anxiety and their impact upon parental perceptions of a child's HRQoL. The removal of the IPQ and STAI items may reduce the length of the PICQoL questionnaire considerably, thus affecting its completion time and enhancing its practicality and clinical utility.

6.11.5 Scoring system

The main choices with a scoring system for HRQoL measures are to aggregate scores from separate dimensions, or to keep them separate (Pal, 1996). Aggregation allows groups to be compared easily, including comparisons of changes in HRQoL over time (Shumaker, Anderson & Czajkowski, 1990). However, it has the disadvantage of missing contradictory trends in dimensions leading to loss of sensitivity; an important change in one dimension may be obscured if a total index score is examined (MacKeigan & Pathak, 1992; Pal, 1996). Disaggregated scores have the advantage of providing descriptive detail about how overall improvement was obtained when making decisions about which therapy to select (MacKeigan & Pathak, 1992). A scoring system that calculated scores for each PICQoL dimension was therefore devised rather than combining scores for each dimension to form an overall score, as in a health profile (MacKeigan & Pathak, 1992). Health profiles and batteries have the advantage in that they provide detailed information about QoL impairments, however, their major limitation is that in comparative studies, arriving at a conclusion about which treatment is superior overall is difficult unless one treatment scores higher than the other on all dimensions (MacKeigan & Pathak, 1992). Factor analytic techniques were utilised to determine weights for the PICQoL items (Streiner & Norman, 1995).

Some HRQoL indices provide both an overall QoL score and separate dimension scores; the score for each dimension should be weighted by its importance or value when a total score is calculated (MacKeigan & Pathak, 1992). Another decision regarding scoring is whether equal weight should be given to each dimension and whether measurement

reflects the importance that each individual puts on different dimensions (Pal, 1996). Some HRQoL measures have been developed for adult patients, which allow individuals to identify aspects of their life considered to be crucial to QoL rather than imposing an external value system on individuals (O'Boyle, McGee & Joyce, 1994). For example, the Schedule for the Evaluation of Individual Quality of Life (SeiQoL) (McGee et al, 1991) and the Patient-Generated Index (PGI) (Ruta et al, 1994). It was not possible to apply this approach to the development of a measure for children under the age of five years, but this approach may be considered in older children who have the cognitive capacity to understand important aspects of their life and how it may be affected by illness. The HRQoL measure is an evaluative measure for a client-specific group; the value of developing an overall total score may be warranted to enable the future comparison of groups receiving differing therapies on the PICU to evaluate effectiveness.

6.11.6 Design strengths

The survey research design in this phase met the research objectives, namely the descriptive evaluation of the psychometric properties of the new HRQoL measure. The evaluation of reliability, validity and practicality addressed three of the four essential requirements in the development of a new outcome measure (Streiner & Norman, 1995). The statistics obtained from the survey also provided an overview of the child's HRQoL and presented possible patterns in the data in terms of significant differences and inter-relationships to form the bases for hypotheses and theories, an additional function of surveys (Polgar & Thomas, 1998). A retrospective approach enabled data to be collected speedily. A total sample in the survey enhanced the generalisability of the research findings. The sample was also of sufficient size to develop a new measure (Kline, 1994).

The population to be covered in this study was widely spread geographically as the PICU is a regional unit; self-administered postal questionnaires seemed an appropriate approach to target this population as this method is cheap and limited funds were available (Moser & Kalton, 1996; Polgar & Thomas, 1998). As the questionnaires were self-administered, the personal influence and bias of the interviewer was minimised (Moser & Kalton, 1996). The closed-response format in the PICQoL questionnaire was tightly structured, allowing responses to be easily encoded and analysed and less time taken to collect responses (Polgar & Thomas, 1998). The response format of the five-point Likert-type questions also allowed a middle 'undecided' response to be recorded.

6.11.7 Design limitations

Approximately 30% of all children admitted to the regional PICU are from the ethnic minorities. The opportunity for parents from ethnic minorities to participate in the survey may have been limited by their inability to read or speak English; the sample comprised only 11% of parents from ethnic minorities. It has been reported that mailed questionnaires can lead to under-representation of ethnic minorities (McColl et al, 1998). The proportion of parents from ethnic minorities participating in this survey might have been enhanced through the use of disproportionate stratified sampling techniques where variable sampling fractions are used (Moser & Kalton, 1996). However, the difficulty in choosing sampling fractions is that fractions that are best for one variable or attribute being studied, say ethnicity, may not be so for another. If one variable predominates in importance, this can govern the sampling fractions, but where no priority exists then the problem of allocation is complex (Moser & Kalton, 1996). While proportionate stratification as a general rule guarantees no loss in precision over simple random sampling; this is not true for disproportionate stratification. An optimum allocation for one variable may yield an allocation that gives for another variable much lower precision than a simple random sample (Moser & Kalton, 1996). Results are also often wanted separately for special sub-groups of the population, domains of study, so it is important to have sufficient sample numbers in each domain. The primary aim of the survey was to evaluate the psychometric properties of the PICQoL questionnaire, and not to explore differences between parental responses according to variables such as sex and ethnicity. However, parental differences in HRQoL perceptions would be valuable to investigate in the future.

The research setting may not be representative of other general PICU's across the UK. Therefore, the external validity or the extent to which the results of an investigation can be generalised to other samples (population validity) or situations (ecological validity), is limited. Other UK general PICU's admit children with similar respiratory illnesses as in this study, but child and parent demographics may differ, such as child and parental ethnic background and parental employment status. This may threaten population validity. The treatment and management of a child's illness by the PICU team may differ in other UK units, medical and nursing expertise and the resources available may also vary, which may affect ecological validity (Polgar & Thomas, 1998).

The sample size in this study was enhanced for certain aspects of the data analysis, namely FA, by merging data from the questionnaires and previous interviews (Phase II). The data was therefore collected at different points in time, which may be considered a bias in the study design. However, further bias was minimised by removing duplicate parents from the survey data, which limited the sample size further.

Response rates to the survey were fair for at least one-parent responders (66.4%); self-administered questionnaires can produce poor response rates. A prospective survey design may have yielded better response rates than a retrospective design, but would have been complex to administer, involving training of ward staff to identify suitable children for the survey, and increasing the cost and length of the study. Recall bias may also have been minimised in a prospective design, as parents would have more recently experienced the PICU admission. However, anecdotal evidence from the Phase I interviews, to develop items for the HRQoL measure, clearly revealed that parents could remember their child's admission to PICU in great detail, even when it was twelve months previously.

Self-administered questionnaires rely upon 100% literacy and a common language; they are inflexible in that responses cannot be clarified; they are inappropriate where spontaneous answers are wanted; responses cannot be treated independently as the respondent can see all the questions before answering any one of them; the researcher cannot be sure that the right person completed the questionnaire; and lastly there is no opportunity to supplement the respondent's answers by observational data which provides useful background material (Moser & Kalton, 1996, Bowling, 1997b). They are prone to respondent and recall biases (Bowling, 1997b; Polgar & Thomas, 1998).

The use of interview-administered questionnaires may minimise response bias, but would require extensive resources, and was therefore not a consideration in this phase. Response bias was minimised in this survey by asking parents to make an explicit assessment of their child's emotional response to their condition following PICU admission. Potential biases in responding to questionnaires include optimising, satisficing, social desirability and faking good, deviation, faking bad, acquiescence, end-aversion, positive skew, halo and framing (Streiner & Norman, 1995).

Satisficing (giving an answer which is satisfactory but not optimal) was minimised by designing the PICQoL questionnaire in a simple format, keeping questions reasonably short and the words easy to understand (Streiner & Norman, 1995). Unfortunately as the questionnaire was retrospective, it was difficult to avoid asking parents how they felt about a situation in the past (e.g. child's PICU admission). Maintaining motivation can also decrease satisficing and may have been a problem in this study as the PICQoL questionnaire, including other measures (IPQ and STAI), was more than 100 items (or 10 pages) (Streiner & Norman, 1995). Social desirability and deviation (tendency to respond to items with deviant responses) was not considered to be a major bias in this study, as the items explored HRQoL subjectively and information sheets stressed that there were no right or wrong answers. Faking good and faking bad was minimised through careful wording of the items.

End-aversion, positive skew and halo effect are biases with Likert scales. End aversion bias (central tendency bias) is the reluctance of some respondents to use extreme categories of a scale, and was seen to some extent in the calculation of the endorsement rates. End-aversion may be minimised by using statements such as 'almost never' instead of 'never'. However this technique was not utilised in this survey so as to encourage respondents who want to reply with absolutes to do so (Streiner & Norman, 1995). A positive skew towards the favourable end of a scale may be a bias producing a ceiling effect (Streiner & Norman, 1995), this can be minimised by ensuring that the 'average' response is not placed in the middle of the Likert scale (Streiner & Norman, 1995). The 'halo effect' was not a bias in this survey, as the parents completed the questionnaire and no judgement was made by an external observer of the child's HRQoL. Framing was not a bias in this survey, as the manner in which a question was posed was not believed to affect the parents' response.
Inter-observer reliability was not assessed in this study; only parental assessments of a child's HRQoL were measured. Testing the inter-observer reliability of the PICQoL questionnaire with clinician assessments may have enhanced the reliability of the PICQoL questionnaire. However, evidence suggests that parental and clinician assessments of a child's HRQoL differ in certain dimensions (Pantell & Lewis, 1987; Rosenbaum & Saigal, 1996; Manificat et al, 1999). Proxy reports of more observable dimension, such as physical functioning and cognition, are more highly correlated with reports from patients themselves (Guyatt et al, 1993). For functional limitations, proxy respondents tend to consider patients more impaired, this is partly true of those proxies with the greatest

contact with the respondent (Rothman et al, 1984). For other sorts of morbidity, patients tend to report the most problems, and then close relatives, and clinicians report the least (Guyatt et al, 1993). Ideally, clinicians should concentrate on ascertainment of reported behaviours and perceptions of patients themselves, and limit inferences they make on the perceptions of caregivers.

Parents from ethnic minorities may differ culturally from sections of the community and this can affect the measurement properties of the HRQoL measure; translation of the PICQoL questionnaire into different languages may have provided an alternative to enhancing representation from ethnic minority groups, but was beyond the scope and available resources. However, the translation of any HRQoL measure is not a simple operation as it is subject to one overriding requirement – equivalence between the source and target version, and subject to two constraints of time and cost (Acquadro et al, 1996). The linguistic validation of an HRQoL measure is also only achieved when the psychometric properties of the translated questionnaire are documented (Nord, 1991; Acquadro et al, 1996). The translation process is a complex iterative process, comprising forward step forward translation of a questionnaire originally developed in a source language into one or more target languages, quality control (quality ratings and back translation), pretest and international harmonisation (comparison of translated questionnaires at an international meeting) (Acquadro et al, 1996).

Some of the data was analysed on the assumption that the data did not come from a normal distribution as illustrated by the frequency distributions of the PICQoL scores, which revealed some skewed distributions on the histograms of PICQoL scores. Therefore, non-parametric statistical methods were used, particularly in the data analysis of test-retest reliability. However, it may have been possible to transform the PICQoL scores of non-normally distributed data and thus utilise more robust parametric statistical tests. Larger samples might also have allowed parametric tests to be conducted.

The assessment of responsiveness to change was not fully addressed in this phase, although the test-retest reliability assessment may give an indication of the responsiveness of the PICQoL questionnaire. Responsiveness refers to a measure's ability to detect change, and will be directly related to the magnitude of the difference in score in patient's who have improved or deteriorated (the signal) and the extent to which patients who have not

changed provide more or less the same scores (the noise) (Guyatt et al, 1993). Comparing the mean change in PICQoL scores for a group of stable children and a group of children whose respiratory illness was known to change may assess responsiveness. The responsiveness of evaluative measures, such as the PICQoL questionnaire, may be compromised by ceiling effects in which patients with the best score may have substantial HRQoL impairment or floor effects in which patients with the worst score may deteriorate further. These effects therefore need consideration in future research to assess the responsiveness of the HRQoL measure.

6.11.8 Advantages of the PICQoL questionnaire over the HUI

Results from the analysis of HUI data revealed that only six utility scores could be calculated. Most parents recorded the last response option in the HUI questionnaire, indicating that the question was not appropriate to the developmental age of their child, suggesting that parents did not find the question appropriate for their child. This finding questions the face and content validity of the HUI measure. Similar difficulties in parental understanding of HUI items have been described (Chapter 2).

The PICQoL questionnaire has several advantages over the HUI measure in the assessment of HRQoL of young children admitted to PICU with a respiratory illness. These advantages relate mainly to how the measure was developed. The PICQoL questionnaire was developed with the intended users of the measure, that is parents of children under the age of five years, admitted to a PICU with a respiratory illness, with some items generated by clinicians. Thus the PICQoL measure comprised important dimensions of HRQoL as identified by parents and clinicians in the population under study. Ideally patients themselves should provide information on HRQoL, however in this study the children were too young and too ill, hence the development of a proxy measure.

The PICQoL questionnaire comprises items that may be classified as generic items and items that are system-specific. Generic HRQoL measures, such as the HUI, are not exhaustive in their assessment of HRQoL and additional disease-specific or dimension-specific information may be required under certain conditions (Gemke & Bonsel, 1996). The advantages and disadvantages of both generic and disease-specific measures are discussed previously (Chapter 1).

The PICU population is particularly heterogeneous with diverse patient groups being admitted (Gemke, 1999). There is also heterogeneity of PICUs with differing treatment options available and expertise (Gemke, 1999). Therefore, the use of a generic measure alone in this PICU population may not have been responsive to the HRQoL changes in this population following treatment. Furthermore, the PICQoL questionnaire was designed to be an evaluative HRQoL measure for use in children following PIC for a respiratory illness. The measure was designed to evaluate changes in HRQoL in this group of children, thus evaluating the effectiveness of PIC. However, it is recognised that further research is needed to define whether a change in score represents a trivial, small but important, moderate, or large improvement or deterioration. One strategy might be to classify children into those who had important improvement as well as those who did not, and examine the changes in the scores in the two groups (Guyatt et al, 1993). Alternatively, one could interpret observed changes in HRQoL measures in terms of elements of those measures that will be familiar to the respondent (e.g. mobility) or determine how scores in HRQoL measures relate to marker states that are familiar and meaningful to clinicians (Guyatt et al, 1993).

6.11.9 Additional items – STAI and IPQ

Parental reports of their children's HRQoL may be significantly affected by their own anxiety and adjustment level (Levi & Drotar, 1998). Therefore, there are good reasons to consider a multi-informant assessment of HRQoL, which includes children's own reports of HRQoL. A measure was therefore included in the PICQoL questionnaire to evaluate parental anxiety levels and their potential impact upon parental perceptions of a child's HRQoL. Results from the PICQoL data (Time 1) revealed that the mean parental STAI score was 41.8, i.e. above a 'normal score' (Marteau & Bekker, 1992). This suggests that parents surveyed in this phase were highly anxious about their child's health and wellbeing, even though they may have been discharged several months previously. Evaluating parental anxiety may identify those parents not coping, which may impact upon future psychological care.

Perception of illness varies on an individual and ethnic level. This is not to say that cultural background determines how its members perceive illness, but that it forms the foundation for defining and responding to illness, e.g. pain sensation (Hutchinson, 1996). Hutchinson (1996) describes that the patient's psychological state, social interaction, physical function

and somatic sensation inform illness perceptions, which in turn inform QoL perceptions. One approach may be to weight items according to the value individuals place on single items within dimensions and/or the overall dimension. A second approach may be to develop an HRQoL measure specific to a particular ethnic group, thus taking into consideration individual experiences of health and illness (Hutchinson, 1996). Alternatively a QoL questionnaire could be developed specifically for a particular ethnic group, however extrapolation of the questionnaire to groups for which they were not intended may not be valid (Hutchinson, 1996). It cannot be assumed that all individuals belonging to a particular ethnic group will respond to illness in the same way; besides life experiences, there are differences in perceptions of illness related to age, sex and class (Hutchinson, 1996).

Findings from the evaluation of parental illness perceptions in this phase identified that parents of children with an acute respiratory illness perceived their child's illness to last for a shorter time than did parents of children with a chronic respiratory illness. These parents also perceived their child's illness to be less controllable and curable than did parents of children with an acute and chronic respiratory illness. This may suggest that parents of acutely ill children perceived their child's illness to be more serious than did those parents of chronically ill children, findings consistent with Marteau and Johnston (1986). However, parents of children with an acute respiratory and other chronic illness perceived their child's illness to be less curable and controllable than parents of children with an acute respiratory illness. Thus suggesting that the type of chronic illness may impact upon parental illness perceptions and possibly coping styles. The presence of an acute respiratory illness may be unfamiliar to a parent of a child with a non-respiratory chronic illness and therefore more threatening; the presence of the other chronic illness might enhance vulnerability.

These findings, however, must be treated with caution in light of the findings from the FA of the IPQ, which did not completely replicate the factors identified by the original scale developers. Although the reliability of the Carer-IPQ appeared promising, the use of confirmatory FA would have been more appropriate to test the hypothesised factor structure of the IPQ in this case. However, confirmatory FA was not performed because of the relatively small sample size employed (Kline, 1994).

6.12 Summary

In summary, the survey conducted in Phase III established the internal consistency and homogeneity, test-retest reliability, criterion validity, construct validity and practicality of the HRQoL measure in a PICU population. The assessment of responsiveness requires future investigation. Generally, statistical findings were within acceptable ranges for establishing reliability and validity, although some responses to PICQoL scores were skewed. Preliminary findings illustrate favourable construct validity yet further research is needed to test constructs in addition to anxiety and illness perceptions. The STAI items correlated well with other items measuring parental anxiety within the PICQoL questionnaire, therefore the removal of the STAI items warrants further consideration. Further research is needed to test the validity of the Carer IPQ in a larger PICU population, as the findings from the FA were inconclusive. The relationship between IPQ scores and PICQoL scores also warrants further investigation.

CHAPTER 7 – Discussion, recommendations and conclusion

7.0 Introduction

This chapter summarises the main findings of the research programme and discusses how the thesis has added to previous knowledge. The research findings are interpreted in the context of the measurement of health outcomes in paediatrics and PIC, with a description of methods to enhance the generalisability and psychometric properties of the HRQoL measure. Recommendations for the future use of the HRQoL measure in the context of research, clinical practice and health policy are also presented.

7.1 Role of HRQoL measures in health policy

This thesis developed and validated an evaluative measure of HRQoL for children under the age of five years following PIC. The use of health and HRQoL outcomes are advocated to measure the benefits of health expenditures and to assess structure and process of health care delivery (Patrick & Erickson, 1996). Cost-benefit, cost-effectiveness, and cost-utility analyses of health interventions may be conducted to compare health outcomes and healthcare costs; the results of these analyses can be used as guidelines for distributing resources to interventions with a lower ration of costs to outcomes (Patrick & Erickson, 1996). Generic HRQoL measures incorporating patient values and generating a single index (preference-based measures) can inform priority-setting policies across very diverse activities (Williams, 1996). Because policy is made at many different levels in the healthcare system (clinical, practice, provider organisation and purchasing organisation), by many different people (e.g. clinicians and managers), serving many different clienteles, there is a role for many different types of HRQoL measures (Williams, 1996).

The evaluative HRQoL measure described in this thesis may inform healthcare policy in a number of ways. It can provide policy-relevant information on the effectiveness of PIC by comparing outcomes in clinical trials, assessing the outcome of new treatments, and evaluating PIC interventions and methods to improve PIC (Steinwachs, Wu & Cagney, 1996; Eiser & Morse, 2001a). The HRQoL measure described may also provide information to inform patients/parents regarding treatment choices and their likely impact on outcomes (Eiser & Morse, 2001a). This can aide an understanding of the parental viewpoint and facilitate improvements in clinical decision-making by informing clinical policy with a description of the consequences of alternative courses of action which can be

shared with patients/parents (Williams, 1996). The evaluation of the quality of medical care, estimation of the health care needs of a population, an understanding of the causes and consequences of the differences in health, and commissioning programmes of care can also be assessed (Speith & Harris, 1996; Eiser & Morse, 2001a). Relevant information can be provided for policy makers facing issues involving benefit design, provider organisation, and payment reform (Steinwachs et al, 1996).

The goal of effectiveness research is to provide the information needed by policy makers, administrators, providers, and patients to improve the provision of healthcare; patient outcome studies can provide insights into the range of opportunities that exist to improve the effectiveness of existing healthcare services (Steinwachs et al, 1996). Health policy debates regarding the role of high-cost technology such as intensive care, policy decisions to cover or not to cover specific procedures, and professional and public perceptions of the appropriate role of the patient in decision making are all likely to change as better and more comprehensive information on outcomes of care become available (Steinwachs et al, 1996).

7.2 Health outcomes in PIC – a summary of the thesis' main findings

Streiner and Norman (1995) recommend a number of stages in the development of an HRQoL measure, including devising items (empirical and/or theory), selecting items (face/content validity, endorsement, homogeneity), and testing the psychometric properties of the measure (reliability, validity, responsiveness and practicality). Desirable properties of HRQoL measures include the assessment of the afore-mentioned psychometric properties (Streiner & Norman, 1995; Jenkinson & McGee, 1997). Fitzpatrick (2000) also describes the properties of precision, interpretability, acceptability (user-centredness) and feasibility (clinical utility). Guyatt, Kirshner and Jaeschke (1992) propose an alternative conceptualization to the requirement of a measure, namely validity and a high ratio of signal to noise. For an evaluative HRQoL measure the signal is the within-subject differences related to true within-subject change, and the noise is the within-subject differences unrelated to true within-subject change; the signal: noise ratio is responsiveness which can be measured using an index of responsiveness (ratio of minimal important difference to standard deviation of changes in stable subjects) (Guyatt et al, 1992).

This thesis demonstrates how parent and clinician interviews, and a systematic review generated items to develop the HRQoL measure; and how the psychometric properties of the HRQoL measure were tested.

7.2.1 A summary of the literature

The availability of evidence describing clinical and cost-effectiveness studies in PIC or suitable measures is limited (Chapter 1). Much of the published literature concerning outcomes of PIC addresses mortality rates or evaluation of health/QoL from a single dimensional perspective, for example, psychological, emotional, physical or cognitive outcomes. Only two studies (three papers) report the measurement of multidimensional health outcomes following PIC (Gemke et al, 1995; Gemke & Bonsel, 1996; Morrison et al, 2000). Few studies consider the child's perspective following PIC; none used outcome measures (Kendrick, 2000; Noyes, 2000; Playfor et al, 2000). Gemke et al (1995) and Gemke and Bonsel (1996) report the most comprehensively conducted studies, but the selected HRQoL measure has its limitations. Children under the age of one year were excluded which represents the largest proportion of children admitted to PICU. Morrison et al (2000) did not use a measure of HRQoL in their study; their choice of measure was also limited by its age applicability.

The author's systematic review demonstrated that few HRQoL measures are available for use in children under the age of five years in a PICU setting (Chapter 2). A new HRQoL measure was indicated for the population under study. None of the measures reviewed were suitable owing to their poor psychometric properties or limited information on their development. The exception being the HUI measure, which was selected as the best available 'gold standard' measure, despite its limitations (Chapter 2).

The author's systematic review was updated in 2001, following the commencement of the final phase of the research programme, and three further generic HRQoL measures were identified: i) TNO-AZL Pre-school Children Quality of Life (TAPQOL) questionnaire (Fekkes et al, 2000); ii) EuroQol (Stolk, Busschbach & Vogels, 2000), and iii) TedQL measures (Lawford et al, 2001). On review, these measures were not suitable for the research programme either for the following reasons.

The TAPQOL measure is for use in children aged 1-5 years and meets the age-range of children studied in this research programme, but complete evidence of its psychometric properties including test-retest reliability and responsiveness is not available. The measure was developed in a Dutch population; differing dimensions of health may be identified in a UK population, but an English-version of the measure is available. Three scales cannot be completed for children aged 1.5 years or older; this represents a significant proportion of children admitted to PICU (Chapter 1). Practicality of the measure is assumed to be reasonable as the measure comprises 43 items, but completion times are not reported.

The EuroQol is a generic QoL measure consisting of two parts: a descriptive system of health (EQ-5D) and a visual analogue scale (EQ_{vas}) (Stolk et al, 2000). The EQ-5D is composed of five single-item attributes each of which is scored at three levels; it can be converted into a weighted health state index ($EQ\text{-}5D_{index}$). This can be used to generate a single outcome measure, which can be used to calculate QALYs. The EuroQol has been well validated in adults but not children. Stolk et al (2000) conclude that the results of their study support the validity (convergent and construct) of a proxy version of the EuroQol to measure QoL in children, but suggest that the $EQ\text{-}5D_{index}$ can be validly used from children aged five years on, and that care should be taken when using the EQ_{vas} in children from 5-10 years.

The TedQL measure is a generic, self-report measure of QoL for children aged 3-8 years developed in the UK. Two versions are reported with the latter version comprising 23 items covering five dimensions. The measure is interviewer-administered using puppets (teddies); completion times are not reported. The measure has been tested in two small-scale studies comprising 36 and 28 children (Lawford et al, 2001). Face and construct validity of the measure is supported, but the internal consistency of the measure is reported as moderate (α=0.60). This measure does not encompass the complete-age range of children in this study and requires an interviewer to administer the measure, which is resource intensive. Limited evidence is available on the psychometric properties of the measure; test-retest reliability and responsiveness are not reported.

Validation studies using HRQoL measures may be classified into stages: Stage I represents studies by the original authors; Stage II represents studies undertaken by other investigators on contrasting samples of people; Stage III studies represent the results of

diverse trials which are combined to provide an overview; and Stage IV studies are comparative validations studies undertaken in different countries (McDowell & Jenkinson, 1996). This research programme represents a Stage I study, but a Stage II study is currently in progress (National Research Register Project: NO436098940, investigator Kay Rushforth). The study is a RCT investigating the clinical effectiveness and cost-effectiveness of nurse-led versus medical-led weaning of children from mechanical ventilation. This classification system may have been useful to apply to the assessment of studies in the systematic review to indicate the level of development of a measure.

7.2.2 Generating the items for the HRQoL measure

Ideally a measure of HRQoL in children should focus on dimensions that are relevant to childhood perceptions of wellbeing, be sensitive to change over time, be sensitive to small changes in health status, measure various health problems, and be simple enough to administer quickly and effectively to children from a wide age range (Stein & Jessop, 1990). Child HRQoL measures must be applicable across all developmental levels and must also include the genetic, biochemical, functional, mental, and prognostic elements of health (Bergner, 1985). This is most critical for assessment of children because their age and development may permit alteration and improvement of many of these elements; the level of any one of the elements may indicate problem areas in health care delivery (Bergner, 1985). Pantell and Lewis (1987) define the ability of a child to fulfil age-related activities, including physical, emotional and social activities, as integral components of a child's health. Child-completed HRQoL measures have been used previously in children as young as four or six years of age (French et al, 1992; Glaser et al, 1997a), and several other child-complete HRQoL measures are reported (See Appendix II). Evidence suggests that children as young as three years of age have a concept of what kind of person they are and how they behave in different situations (Eiser & Morse, 2001b; Lawford et al, 2001).

As the age-range of the children in this study was less than five years, a child-completed HRQoL measure was not feasible to develop and was inappropriate; some children were too sick to contribute to its development. The views of parents and clinicians were therefore elicited to develop and complete the measure, as recommended by Streiner and Norman (1995) (Chapters 3-4). The use of proxy-respondents to evaluate HRQoL outcomes has been criticised; the parent-report may provide a substitute for a children's QoL at a group level, but large differences can exist in proxy agreement at the individual

child-parent level (Theunisson et al, 1998). It has been previously reported that proxy-respondents less adequately measure the more subjective dimensions of health status than the more objective domains including functional capacity (Glaser et al, 1997a). Theunisson et al (1998) suggest that until more conclusive evidence is obtained indicating that one informant is more reliable, information should be collected from multiple informants.

Only parents (mothers or fathers or both) or the child's main carer recorded an assessment of the child's HRQoL using the PICQoL questionnaire in this study (Chapters 5-6). Assessment by clinicians was not performed. However, clinicians contributed to the development of items to strengthen the clinical utility and feasibility of the HRQoL measure (Chapter 4), and to promote its clinical application in routine clinical practice (Drotar et al, 1998a). Employing clinicians to rate the child's HRQoL in addition to parents could have provided a multi-informant perspective, and contributed to establishing concurrent validity. Clinician ratings of HRQoL are recommended for future research.

Eiser and Morse (2001a) identify many studies of HRQoL with children, which include both mothers and fathers in their samples, although the number of fathers tended to be small. This research programme included the views of fathers, in addition to mothers, in the development and testing of the HRQoL measure (Chapters 5-6). Fathers represented 22.2% (n=6) of respondents in Phase I (Chapter 3), and their views were considered in generating HRQoL items for the measure and exploring meanings (Drotar et al, 1998b). Sixteen (22.2%) fathers also participated in a cross-sectional survey to test PICQoL items for importance, agreement and dimensionality (Chapter 5). Forty-one (33.9%) fathers also participated in a cross-sectional survey to test the reliability, validity and practicality of the PICQoL questionnaire (Chapter 6). This research programme therefore represents the largest published child HRQoL study involving fathers or male carers. Further data analysis to determine the similarities and differences between PICQoL scores, and STAI and IPQ scores for mother and father pairs, and mothers versus fathers may add new knowledge to the ability of fathers as well as mothers to act as suitable proxies for their children. This analysis is recommended for future research.

Parents described the concept of health and QoL as possessing similar dimensions, including emotional, behavioural, cognitive, social, physical (nutrition, appearance) and family characteristics (Chapter 3). These dimensions are similar to those described by

Fitzpatrick et al (1998b) for patient-based outcome measures, that is a multidimensional concept including physical function; symptoms; global judgements of health; psychological wellbeing; social wellbeing; cognitive functioning; role activities; personal constructs; and satisfaction with care. Parents perceived the difference between health and QoL as health forming a component of a child's QoL. Parents perceived physical aspects as the most important attribute in the theme of global health, and emotional aspects as the most important attribute in the theme of global good QoL. These findings are partially consistent with those described by Eiser and Morse (2001a), who reported greater emphasis being placed on mental, compared with physical functioning when rating QoL, and greater weight being placed on physical functioning when rating health status (Eiser & Morse, 2001a).

Clinicians identified similar dimensions of health and QoL to those perceived by parents, particularly relating to the impact of respiratory symptoms on the child's level of functioning and wellbeing (Chapter 4). Clinicians often relied upon the subjective assessment of a child's HRQoL through consultation with parents; few objective respiratory outcome measures are routinely used in clinical practice. The subjective nature of HRQoL measures is a source of some unease among investigators. However, the heterogeneity of some patient populations and an inability to identify, let alone control, all variables that influence disease progression have forced broader measurement tolerances to be accepted in clinical medicine (Schipper et al, 1996). Measures of HRQoL use patients as their own internal controls, and can be used without norms (Schipper et al, 1996). With this approach, the critical HRQoL value is not the score a patient provides, but rather the change in that patient's score over time (Schipper et al, 1996). When making comparisons of groups of patients, the central issue is not whether the overall score in one group is better than the other, but rather whether the change in scores observed over time is different in each group (Schipper et al, 1996). In relatively homogenous populations it is probably reasonable to look at differences in raw scores, but many of the problems associated with comparing people of different social, economic, and cultural milieus are circumvented when change in score within patients becomes the focus of the examination (Schipper et al, 1996).

Psychological factors have an impact on HRQoL perceptions and the ones most studied include anxiety, depression, and fear (Schipper et al, 1996). Parental anxiety was a

prominent theme emerging from parental data in this study (Chapter 3), and a validated measure of anxiety, the short-form STAI measure was incorporated within the HRQoL measure to evaluate levels of parental anxiety regarding their child's PICU illness (Chapter 5). Parental illness perceptions were also a pronounced theme, and a theoretical framework of cognitive illness representations was applied to the analysis of data (Chapter 3). The Carer-version of the IPQ was adapted for use with parents of critically ill children and incorporated within the HRQoL measure to evaluate parental cognitive illness perceptions and to explore potential relationships with parental perceptions of HRQoL.

Eiser and Morse (2001a) state that parents will be influenced by the development of other children they know (their own or their friends), their expectations and hopes for their child, additional life stresses, and their own mental health. They recommend that research should focus on determination of how proxy mental health influence ratings of the child's QoL, and to clarify how parent mental health and perceptions of the child's disease influence QoL over time (Eiser & Morse, 2001a). This is relevant to issues concerning how parenting practices and family organisation can subsequently affect the child's QoL. The inclusion of the STAI and IPQ measures attempted to address these recommendations.

7.2.3 Scaling responses in the HRQoL measure

The PICQoL questionnaire was developed to reflect all of the dimensions of health described in the literature and those described by parents and clinicians (Chapter 5). The questionnaire contained two summary items asking parents to rate their child's overall health and level of breathing. Summary items have the advantage of brevity and validity, but their disadvantage is an inability to reveal contradictory trends in different dimensions of health (Fitzpatrick et al, 1998a). However, inviting a respondent to summarise their health, in this case a child's health, offers a potential method for weighting up the significance of such contradictory trends (Fitzpatrick et al, 1998a). Summary items were not used in isolation in the HRQoL measure; more detailed information on a child's health and HRQoL was obtained.

The PICQoL questionnaire also contained two transition items which asked parents to compare their child's overall health, and level of breathing, with a specific point in time, namely before PICU admission. Fitzpatrick et al (1998a) provide evidence of studies illustrating transition items to have good validity by producing scores consistent with

independent evidence of the direction of change in health experienced by respondents between separate assessments. However, sometimes respondents report poorer health states than actually experienced so that the degree of improvement is exaggerated; some respondents may also be unduly influenced by their current health state when asked to compare current with past health (Fitzpatrick et al, 1998a).

Response options in an evaluative HRQoL measure should have sufficient graduations to register change, and most scale-developers choose a seven-point scale (Guyatt et al, 1992; Juniper et al, 1996). A five-point response option was chosen for most of the items in the PICQoL questionnaire. Streiner and Norman (1995) state that the use of five categories can reduce final reliability by about 12%; they suggest that the minimum number of categories used by raters should be in the region of five to seven. There is, however, good evidence that, in a wide variety of tasks, people are unable to discriminate much beyond seven levels (Streiner & Norman, 1995).

A scoring system was devised for the PICQoL questionnaire calculating scores for each dimension rather than an overall total score to allow for important changes in any one dimension to be identified (Chapter 6). This is particularly important when making decisions about which treatment to select (MacKeigan & Pathak, 1992). Factor analysis was utilised to develop weights for certain PICQoL items, although ideally it is recognised in paediatrics that it is important to confront how both parents and children value aspects of health in order to drive empirically a weighting system (Pantell & Lewis, 1987). Most models of health in children do not assign particular weights to various influential factors, factors are implicitly weighted differently for different individuals and as they change over time (Pantell & Lewis, 1987). Ipsative methods place the individual reporter's perspective at the centre of the HRQoL assessment. One ipsative approach, the PGI, may be especially useful as a family-centred approach to HRQoL assessment that enhances clinicians' understanding of how families integrate patients' illness experience into their lives. As the patient selects the areas of concern, the PGI may be used to gather developmentally appropriate concerns of children (Jacobson & Fried, 1998)

Ten PICQoL scores were developed reflecting general health, comparison of general health, positive milestones, negative emotions, impact on family, parental worries, general breathing, comparison of general breathing, respiratory symptoms and respiratory

limitations; a further five scores were also calculated from the IPQ and STAI items (Chapter 6). The PICQoL scores reflect dimensions of general health (global judgements of health), physical functioning, cognitive functioning, emotional functioning, social functioning, psychological functioning, symptoms (respiratory), and family functioning. These scores reflect the dimensions of HRQoL described by others (Speith and Harris, 1996; Ware, 1987; Spilker, 1996; Fitzpatrick et al, 1998a).

7.2.4 Selecting the items for the HRQoL measure

The redundancy of items within the PICQoL questionnaire was determined by the item failing to meet one of two criteria: an endorsement range of 0.2-0.8, or an ITC above 0.2 (Streiner & Norman, 1995). Most items met one of these two criteria, but some demographic items were removed from the questionnaire because they were judged as clinically irrelevant by expert clinicians in the RAG. A third criterion of FA was also applied. The final HRQoL measure, tested in Phase III, comprised 66 HRQoL items with additional items on child and parent demographics, IPQ and STAI, and questionnaire evaluation (Chapter 6). Bowling (1997a) states that items that deliberately tap different dimensions within a measure cannot be expected to necessarily have high item-item or ITCs, so FA should be used to identify the separate factors within the measure. Exploratory factor analytic techniques (PAF and PCA) were utilised in Phases II and III to assess the dimensionality of the measure by grouping together variables that correlated, and to identify redundant items. In PAF only the variance, which is common to or shared by items, is analysed, so an attempt is made to exclude unique variance (specific variance plus error variance) from the analysis; in PCA all the variance of an item is analysed including its unique variance (Bryman & Cramer, 2001). Data from Phase II and III samples were pooled to increase the sample size for the purposes of FA (n=153); however, pooling data may have obscured differences between diverse groups rather than illuminating them (Tabachnick & Fidell, 2001). The sample size was sufficient to conduct FA; there should be more participants than items (Bryman & Cramer, 2001).

The graphical scree test was utilised to determine the number of factors to keep. Orthogonal (varimax) rotation was utilised in the analysis to increase the interpretability of the factors. The advantage of orthogonal rotation is that the information the factors provide is not redundant, since a persons' score on one factor is unrelated to his/her score in another; however, the disadvantage is that factors may have been forced to be unrelated

when in real life they may be related. This may be less likely with oblique rotation (Bryman & Cramer, 2001). Items or variables which correlated less than 0.3 with a factor were omitted from consideration, as is the convention, since they account for less than 9% of the variance and so are not very important (Bryman & Cramer, 2001). The criterion of the correlation above which no item correlates more highly with more than one factor was also utilised; many researchers ignore this second criterion and emphasise all loadings in excess of 0.3 regardless of whether any items are thereby implicated in more than one factor (Bryman & Cramer, 2001). Both criteria were used in this study, thus strengthening the rigour of the analyses.

Exploratory FA was utilised to identify relationships between the various items examined without determining the extent to which the results fitted a particular model (Bryman & Cramer, 2001). The FA did reveal structure, but further analysis is warranted in a larger sample using confirmatory factor analytic techniques to confirm the structure for PICQoL and IPQ items. Confirmatory FA compares the solution found against a hypothetical one (Bryman & Cramer, 2001). Fitzpatrick (2000) suggests that regression may be used to identify the subset of items that capture the most variance of an underlying construct rather than relying on factor analytic techniques to identify items with sufficient consistency to each other as to produce the scales of a measure (Fitzpatrick, 2000). Other methods of generating items warrant consideration, such as the 'impact method' whereby items are selected on the basis of the importance to patients with importance determined by the product of patient's ratings of importance and frequency of items (Fitzpatrick, 2000). Such techniques are reported in the development of PGIs whereby aspects of life considered important by patients are evaluated and include the relative value of the different aspects as determined by the patient (Jenkinson & McGee, 1998).

7.2.5 Testing the psychometric properties of the HRQoL measure

7.2.5.1 Reliability

Reliability concerns the extent to which a measuring procedure yields the same results on independent repeated trials under the same conditions (Guyatt et al, 1987; Bowling, 1995). The testing of reliability and validity is dependent upon the purpose of the measure (McDowell & Jenkinson, 1996). The HRQoL measure in this study is an evaluative one, internal consistency (measurement of the same concept by different scale items) and item-response statistics have been described as an appropriate approach to the testing of

reliability in an evaluative measure (Bowling, 1995; McDowell & Jenkinson, 1996). Cronbach's alpha statistic and ITCs were used to calculate the internal consistency of the HRQoL measure by identifying the extent to which items on a scale were tapping a single underlying construct demonstrated by a high level of ITC (Jenkinson & McGee, 1997 & 1998). Cronbach's alpha coefficients ranged from 0.76-0.90 (Phase II) and 0.83-0.87 (Phase III) for PICQoL and STAI items suggesting high internal reliability. However, some alpha coefficients were above the recommended range of 0.7-0.9, in particular the respiratory symptom items in Phase II and III (0.92 and 0.93 respectively). If alpha coefficients are too high, it may suggest that some items are unnecessary and the content validity may be poor (Streiner & Norman, 1995). Alpha coefficients were however low for IPQ items (0.57 in both phases) suggesting that the items do not come from the same conceptual domain (Bowling, 1995). The ITCs (Phase III) revealed that all items correlated with their total scores above 0.2, suggesting homogeneity of the PICQoL questionnaire (Streiner & Norman, 1995).

The assessment of test-retest reliability also determines whether a measure is reliable over time by administering the measure on two occasions separated by a few days (Jenkinson & McGee, 1998). In test-retest reliability it may be difficult to distinguish measurement error from real changes in HRQoL (Fitzpatrick et al, 1992). A more precise definition describes the reliability of a single observation as the ratio of the variance attributable to true differences among patients, to the total variance (the sum of the variance due to true differences and the variance due to random errors of measurement, assuming errors to be independent of the measurement themselves) (Guyatt et al, 1987).

Pearson's correlation coefficient can be used to quantify reliability but fails to take into account variability in results attributable to systematic, as opposed to random, differences in test scores with multiple applications (Guyatt et al, 1987). The ICC, which reflects both systematic and random differences in test scores, is generally accepted as the preferable method of assessing reliability (Guyatt et al, 1997), and is reported in previous studies (Eiser & Morse, 2001a). It was not appropriate to calculate the ICC in this study, as there were two measurements per respondent, with Time 1 being more recent to the PICU admission than Time 2, rather than two different observers. The HRQoL measure in this study showed good test-retest reliability and a high level of consistency in response (Bowling, 1995). Pearson correlation coefficients ranged from 0.577-0.822 (Spearman's

rho 0.510-0.773), and were significant at the 0.01 level. However, it is difficult to interpret whether small correlations for some PICQoL items represented unreliability of the HRQoL measure or reflected real changes in the child's health status, or whether the parent's familiarity with the questionnaire led to changes in their responses (Jenkinson & McGee, 1998). The inclusion of a 'change' question, such *as 'Has your child's HRQoL changed since completing the questionnaire last time?'* or *'Has your child's breathing changed since completing the questionnaire last time?'* could have indicated those children whose HRQoL or breathing had not changed. Thus facilitating the assessment of test-retest reliability.

7.2.5.2 Validity

A valid assessment is one that measures what it claims to measure (Jenkinson & McGee, 1998). Four aspects of validity are assessed for any properly constructed questionnaire, namely, face, content, criterion and construct validity (Jenkinson & McGee, 1998). Evaluative measures should be examined using construct validation techniques such as convergent agreement with equivalent methods, and sensitivity to change or responsiveness (McDowell & Jenkinson, 1996). The latter is often indicated by an effect size statistic, comparing scores before and after an intervention that is expected to alter the quantity being measured (McDowell & Jenkinson, 1996).

Face validity examines whether a measure appears to be measuring what it is intended to measure and content validity examines the extent to which the domain of interest is comprehensively sampled by the items, or questions, in the measure (Guyatt et al, 1993). Face validity of the PICQoL questionnaire was supported by parents who completed the questionnaire acknowledging that the PICQoL items were appropriate to the measurement of HRQoL outcomes post PIC, and that the items were easily understood (Jenkinson & McGee, 1998). Content validity of the measure was supported via expert (RAG) review with clinicians being involved in the generation of PICQoL items, and ensuring that the measure tapped all relevant concepts of the attribute of HRQoL (Bowling, 1995; Jenkinson & McGee, 1998). The weighting of items in the calculation of PICQoL scores was determined statistically via factor analytic techniques rather than asking parents or clinicians to list the items in order of priority to reflect their perceived level of difficulty of the health problem in question (Jenkinson & McGee, 1998). A review of the items by an

expert panel, RAG members, also confirmed the content validity of the measure, and is a minimum prerequisite for acceptance of a measure (Streiner & Norman, 1995).

Validity testing is all about making inferences; if important aspects of the outcome are missed by the measure, then it is likely that some inferences will be proved to be wrong; the inferences and not the measure is therefore invalid (Streiner & Norman, 1995). If a measure has high content validity, then the broader are the inferences that can be validly drawn about the person under a variety of conditions and different situations (Streiner & Norman, 1995). However, difficulties can occur when tapping a behaviour, disorder or trait that is relatively heterogenous, like PIC illnesses; it is quite conceivable that the measure will have low internal consistency as not all patients with one particular PIC symptom or trait will exhibit other similar PIC symptoms or traits (Streiner & Norman, 1995). The internal consistency of the measure could be increased by eliminating items, which are not highly correlated with each other or the total score. However, if this were to happen, the measure would end up tapping only one aspect of the PIC illness, reflecting a very low content validity (Streiner & Norman, 1995). Under such circumstances it is better to sacrifice internal consistency for content validity, as the ultimate aim of the measure is inferential, which depends more on content validity than internal consistency (Streiner & Norman, 1995). Important aspects of HRQoL identified by parents included dimensions of HRQoL described in the literature, in addition to respiratory-specific consequences such as frequency and severity of symptoms and the impact of these symptoms on the child and family life; items were developed to reflect these areas, ensuring high content validity.

Criterion validity was assessed using the HUI measure. The modification of the HUI measure in this study, owing to its poor face and content validity, may have affected its reliability and validity with the further assessment of the psychometric properties of the modified version being required (Cheater, 1998). Significant moderate correlations at the 0.1% level were found between the HUI general health item and PICQoL general health score and between the PICQoL positive milestones and negative emotion scores and HUI II/III attributes of mobility, cognition, self-care, speech, ambulation, dexterity and emotion. This demonstrates satisfactory criterion validity. Moderate correlations were found as specified before the study, thus proving criterion validity of the HRQoL measure (McDowell & Jenkinson, 1998). Another form of criterion validity is discriminant validity, whereby the HRQoL measure should not correlate with dissimilar, unrelated variables

(Streiner & Norman, 1995). Discriminant validity of the PICQoL questionnaire was not tested in this study, but is a recommendation for future research.

Construct validity refers to the ability of a measure to confirm expected hypotheses (Jenkinson & McGee, 1997). Assessment of the construct validity of the HRQoL measure was tested by exploring the statistical relationship between the child's illness type with PICQoL scores relating to parental anxiety and parental illness perceptions including the impact of the child's illness upon family life; that is confirming 'a priori' hypotheses (Pham & Klaassen, 2000). Construct validity proved promising with only two 'a priori' hypotheses not confirmed in the expected direction. The rationale for the chosen hypotheses was given and the constructs were clearly defined (McDowell & Jenkinson, 1996).

7.2.5.3 Responsiveness

It is essential that evaluative measures are able to detect change and the level of this change is interpretable in some way; the sensitivity to change or responsiveness of a measure is a very important criterion to consider when selecting measures (Jenkinson & McGee, 1998). However, measures may be insensitive to change in QoL for several reasons. For example, generic measures may include items not relevant to a particular disease or treatment group; measures may include items that assess areas that are relatively static or not a feasible target of the health care intervention; or measures may be subject to ceiling and floor effects (Fitzpatrick et al, 1992). For patients with a very poor QoL who obtain minimum scores before treatment there may be no scope to register any further deterioration (floor effect), conversely in patients with an excellent QoL who obtain maximum scores before treatment there may be no scope to register any further improvement (ceiling effect) (Fitzpatrick et al, 1992; Jenkinson & McGee, 1997). Several statistical methods of assessing change have been proposed, including the calculation of the effect size statistic, standardised response mean (SRM) or responsiveness index; the statistical nature of these tests has been identified as a potential problem (Jenkinson & McGee, 1998). However, other attempts have been made to make changes interpretable, including asking patients themselves whether a level of change was unimportant, minimally important or of greater importance, when a change is reported (Juniper et al, 1994).

The distribution of scores revealed no PICQoL scores with a maximum or minimum value, suggesting that there may be scope for an improvement or deterioration in scores over time. The assessment of the sensitivity to change of the PICQoL questionnaire is recommended for future research. One method could be to ask a 'change' question as described previously in a test-retest questionnaire. A responsiveness index could be calculated for those children whose HRQoL or breathing was reported to change since the last administration of the PICQoL questionnaire. Alternatively, responsiveness could also be assessed by comparing within person standard deviation to the change in score observed after an intervention of known efficacy (Guyatt et al, 1987). However, care is needed to choose an appropriate intervention. If a poor choice of intervention is made, and the treatment does not change patient status (or produces a change which is clinically unimportant), the responsiveness will be underestimated. To the extent that the intervention produces improvement which is greater than the smallest clinically important difference, an inflated estimate of responsiveness will be obtained (Guyatt et al, 1987).

7.2.5.4 Practicality

Measures of HRQoL are most practical for use in clinical trials and formal evaluation studies, where they are used alongside other information about patients, treatments, and outcomes to address fairly precise questions (Fletcher et al, 1992). For regular use in clinical care or clinical audit the more detailed and comprehensive HRQoL measures are both impractical to administer and process and hard for healthcare professionals to interpret and incorporate into decision making (Fletcher et al, 1992). One of the essential criteria of an HRQoL outcome measure is thus its practicality (Streiner & Norman, 1995).

The practicality of the PICQoL questionnaire was assessed via an item on ease of completion; 70.2% (n=85) of parents (Phase III) found the questionnaire 'very easy' or 'quite easy' to complete. The mean completion time for the questionnaire (Time 1) was 22.4 minutes (range 4-60 minutes), suggesting adequate practicality. Practicality was also assessed via the survey response rate in Phase III, which was 66.4% for at least one-parent responders. A response rate of 60-69% is reported as acceptable (McColl et al, 2001), but others recommend a minimum standard of 75% (Fowler, 1993). Using incentives, monetary or material, at the time of response could have enhanced the response rate (McColl et al, 2001). Motivating respondents to complete a questionnaire is also dependent on the subject matter, the interest of the respondents in the project, and what kind of prior

contact has been made with the respondents, such as methods of pre-notification (Fowler, 1996; McColl et al, 2001). Measures of HRQoL also need to be appropriate to the target population and setting (Cheater, 1998). The PICQoL questionnaire scores highly for appropriateness as it has been tested in the target population selected for the research programme, namely parents of children who received PIC.

7.2.5.5 Precision

Precision of measurement of the outcomes in the relevant range of assessment is critical to the success of an intervention, evaluation and should not be confused with reliability (Kessler & Mroczek, 1996). The HRQoL items in some outcome measures, while quite appropriate for a general population, may be 'too easy' for a patient population, resulting in an inadequate discrimination in the range of the dimension where variation exists (Kessler & Mroczek, 1996). Item response theory methods can be utilised to create measures, which are designed to have precision in a particular range of an outcome's overall latent distribution (Kesler & Mroczek, 1996). Item response theory is a different theory of test construction to generalisability theory, which has underpinned most test construction and theory since its introduction (Streiner & Norman, 1995).

Streiner and Norman (1995) state that the assumptions that generalisability theory makes about items and tests are relatively 'weak', so that the theory is appropriate in most situations, and this is one reason for its popularity. The essence of the theory is the recognition that in any measurement situation there are multiple, infact infinite sources of error variance; an important goal of measurement is to attempt to identify, measure and thereby find strategies to reduce the influences of these sources on the measurement in question (Streiner & Norman, 1995). Generalisability theory attempts to do this by combining all sources of variability in a single study, using all the data to estimate the variance between respondents and the various components of error variance, there are however, a number of limitations to generalisability theory (Streiner & Norman, 1995). Item and scale statistics apply only to the people who took the test; if the measure is to be administered to people who are different in some way it is often necessary to re-establish its psychometric properties. It is also extremely difficult to compare a person's scores on two or more different tests; total scores are usually converted to *z* scores (Streiner & Norman, 1995). Also, it is assumed that the error of measurement is the same at the high end of the scale as in the middle or at the low end (known as homoscedasticity), however,

errors tend to be smaller near the ends of the range of possible test scores where floor and ceiling effects come into play (Streiner & Norman, 1995). Lastly, if a person responds to 50% of items in a positive direction, all that can be said in traditional test theory is that the probability of that person responding positively to any given item is 50%; the assumption is that all of the items have equal strength (Streiner & Norman, 1995). In fact, it is impossible to predict how a person will respond on any given item if the items differ in their propensity to tap the attribute (Streiner & Norman, 1995).

Another set of concerns with classic test theory, upon which the reliability coefficient is based, is that the scores on a measure depend on how much of the trait the people in the sample have, while 'how much they have' depends on the norms of the scale; thus the measure's characteristics change as different groups are tested and the groups' characteristics change as different tests are used (Streiner & Norman, 1995). Item response theory has been proposed to rectify these short-comings, and is based on two hard assumptions: that the data are unidimensional (items tap only one trait or ability); and the probability of answering any item in a positive direction (reflecting more of the trait) is unrelated to the probability of answering any other item positively for people with the same amount of the trait (a property called 'local independence') (Streiner & Norman, 1995). Item characteristic curves can be computed to describe the relationship between a person's performance on any item and the underlying trait using one of three 'models', the simplest being the one-parameter model or Rasch model (Streiner & Norman, 1995). Item-response theory warrants further investigation in this research programme, but may not be feasible as the mechanics of deriving the item characteristic curves involves a large number of participants, a minimum of 200 for estimating a one-parameter model (Streiner & Norman, 1995).

7.2.5.6 Interpretability

Measures of HRQoL should also provide clinically relevant information (Pham & Klaassen, 2000). Interpretability has only recently emerged as an important issue in HRQoL assessment; it raises the fundamental question of how meaningful scores of HRQoL are (Fitzpatrick, 2000). It has frequently been observed that a major barrier to the more widespread use of HRQoL scales in evaluative research is that they lack the intuitive meaning and familiarity to clinicians that are obtained with conventional measures such as blood pressure and temperature (Fitzpatrick, 2000). In studies with traditional outcomes

there is usually a consensus on what constitutes a meaningful clinical effect; as yet there is no similar direct interpretation of HRQoL scores, partly because of the limited experience of these measures in everyday clinical practice and clinical trials (Fletcher et al, 1992).

For an evaluative measure, one approach described is to examine the changes in scores in two groups of patients, those who showed important improvement and those who did not (Guyatt et al, 1993). Observed changes in HRQoL measures may be interpreted in terms of elements of those measures that will be familiar to readers or how scores in HRQoL measures relate to marker states that are familiar and meaningful to clinicians (Guyatt et al, 1996). Another approach is to identify minimal clinically important differences in score that might realistically be perceived as important by the patient or lead to a change of management (Juniper et al, 1994). For measures that present response options as seven-point scales, small, medium, and large effects correspond to average changes of approximately 0.5, 1.0 and >1.0 per question (Juniper et al, 1994). Other methods for interpreting change scores are primarily statistical, and include the calculation of the effect size statistic, or SRM, or responsiveness index, as highlighted previously (Jenkinson & McGee, 1998). One method of interpretation that can be adopted for generic HRQoL measures is the comparison of results before and after treatment with norms for the general population (Jenkinson & McGee, 1998). However, the use of 'population norms' to interpret effects of treatment has its pitfalls and limitations (Fletcher et al, 1992). While it is also possible to find a statistically significant difference in scores on a questionnaire before and after treatment, it need not be all clear that the difference means very much either to clinicians or patients (Jenkinson & McGee, 1998).

In this study, the PICQoL scores were transformed to a 0-100 scale so that a high score represented better HRQoL and a low score reflected worse HRQoL. The mean PICQoL scores for all ten scores ranged from 32.4 – 85.1 (Time 1). A classification system is yet to be developed to aid interpretation of the PICQoL scores, such as a range of scores to illustrate 'excellent HRQoL' or 'poor HRQoL'. It may be helpful to clinicians to develop a guide to the interpretation of very high or low scores, as described by the SF-36 developers (Jenkinson & McGee, 1998). For example in the physical functioning dimension of the SF-36, an adult generic HRQoL measure, a poor score represents 'limited a lot in performing activities including bathing and dressing', and a high score represents 'performs all types of physical activities without limitations due to health' (Jenkinson & McGee, 1998).

Similar levels of functioning are described for the HUI scores (Torrance et al, 1996). A low PICQoL score for the general health dimension may thus reflect the parental belief that a 'child's health is poor and likely to deteriorate', whereas a high score may reflect the parental belief that a 'child's health and HRQoL is excellent'. This will assist clinicians in identifying those children whose HRQoL outcomes have changed over time.

7.2.5.7 Acceptability or user-centredness

Measures should be acceptable to patients; if measures are not minimally acceptable to patients there is a real concern that response rates and rates of missing responses are increased thereby jeopardizing the interpretation of HRQoL outcomes in relation to interventions and possibly introducing bias (Greenhalgh et al, 1998; Fitzpatrick, 2000). One approach to improving the appropriateness of HRQoL measures is to use measures that let patients select the dimensions of most concern, as in PGIs; thus the HRQoL scores will vary from patient to patient and scores can be assessed over time (Fitzpatrick et al, 1992). In general, evidence of acceptability should be examined at the design stage, with the most direct and easy evidence being the length of the questionnaire and the response rates of questionnaires (Fitzpatrick et al, 1998a). Parents were asked in Phase II to comment on questionnaire design, including content relevance; one item was omitted from the measure based upon this assessment, as some parents found it difficult to complete an item assessing anxiety levels in a very young child.

7.2.5.8 Feasibility or clinical utility

Feasibility is the complementary issue to acceptability and practicality and draws attention to the possibility that HRQoL measures may differentially disrupt clinical care and impose burdens on staff (Fitzpatrick, 2000). Measures used in clinical trials are almost invariably completed by patients during routine care and methods of assessing HRQoL that are more time-consuming and require more effort and training to collect and process may be less likely to be successfully integrated into many clinical trials, thus jeopardizing trial conduct and disrupting clinical care (Fitzpatrick et al, 1998a; Fitzpatrick, 2000). An obvious example is the additional staff effort and costs involved in personally administering questionnaires over postal delivery (Fitzpatrick et al, 1998a). To a lesser extent, the length and complexity of a measure are an additional component; certainly it may require additional staff time to assist and explain how more complex questionnaires are to be filled out by patients (Fitzpatrick et al, 1998a). Some measures have been shortened to produce

more acceptable and feasible versions e.g. SF-36 shortened to SF-12, however, brevity may mean that potentially important information about patients' experiences is missed and increased acceptability is achieved at the risk of diminished content validity (Fletcher et al, 1992; Fitzpatrick 2000). The validity and responsiveness of shorter measures needs to be studied, and may be a consideration for future research with the PICQoL questionnaire (Fletcher et al, 1992; Fitzpatrick, 2000).

A related component of feasibility is time required to train staff to use a measure, with questionnaires for self-completion imposing the least burden in this respect; where measures do require interviewer-administration, training needs can vary according to the complexity of the tasks (Fitzpatrick et al, 1998a). The PICQoL questionnaire is designed as a proxy-completed measure and it is thus anticipated that staff would need minimal training to administer the PICQoL questionnaire, if they chose to administer it in a clinical setting. The PICQoL questionnaire, however, can be administered by postal delivery to minimise staff effort and time spent training; this is also a relatively low cost and efficient way to collect information (Gotay, 1996).

It is sometimes thought that more complex scoring systems reduce feasibility compared to simple scores, but this element is unlikely to be a major component of burden to staff with computer programmes universally used to process such data (Fitzpatrick et al, 1998a). The PICQoL data was entered into a widely available statistical data analysis programme (SPSS 10.0) and the scores were calculated using syntax commands. However, a manual describing the development and testing, administration and interpretation of the PICQoL questionnaire, such as that described for the CHQ, is required and recommended (McDowell & Jenkinson, 1996). In the context of the future use of the PICQoL questionnaire in clinical trials, it will be necessary to provide administrative support with data collection, processing and analysis; this will need consideration in any future funding applications. Positive attitudes by staff and acceptance of the value of the PICQoL questionnaire can also make a substantial difference to ultimate acceptability of the questionnaire by patients (Fitzpatrick et al, 1998a).

7.3 Research design and methods

The advantages and disadvantages of the overall research design and methods employed are presented in detail in each chapter. The data collection strategy was driven by the

research purpose, practical considerations, as well as by concerns about data quality (Fowler, 1996; Mays and Pope, 1996b).

7.3.1 Enhancing the generalisability and psychometric properties of the HRQoL measure

Although the scale-development approach described by Streiner and Norman (1995) was followed, a number of factors limited the generalisability of the research findings, and additional approaches could have enhanced the reliability and validity of the HRQoL measure.

7.3.1.1 Alternative methods

The research programme employed a mixture of quantitative and qualitative research methods to develop and validate the HRQoL measure. The research design may have been enhanced by utilising standard physical, psychological, or neurological tests to measure patient abilities that are important aspects of QoL (Gotay, 1996). Appropriate tests depend on the disease, treatment, patient population, and study questions, and some tests may even be administered in the context of patient care (Gotay, 1996). Several tests are available for paediatric populations such as tests to assess pain, cognitive functioning, play performance, and these may have complemented HRQoL information obtained from the PICQoL questionnaire and contributed to the assessment of validity of the HRQoL measure.

Ideally, assessment of HRQoL outcomes should be done outside the hospital setting as patients' problems are rated less severe outside their home (Ebbs et al, 1989). This was possible for parents interviewed and surveyed retrospectively, but not for prospective parents who were interviewed in the PICU setting. Ideally, assessments should be conducted pre-, during and post-treatment (Ebbs et al, 1989). An assessment of the child's pre-PICU health status and wellbeing was assessed via relevant items in the PICQoL questionnaire, but as described previously, the majority of children admitted to PICU are emergency admissions, so it was not possible to target these children pre-PICU admission. However, children who are admitted electively for post-operative care may be targeted pre-PICU admission.

The interviews conducted with parents (Phase I and II) and the postal survey (Phase III) were in retrospect of the child's PICU episode, recall bias might have been a problem with some parents finding it difficult to recollect past events. Prospective interviews in Phase II

aimed to reduce bias owing to recall and memory distortions. A timeframe of the previous two weeks was utilised for questions relating to the child's current health and wellbeing to minimise memory distortions (Fowler, 1996). A further limitation may have been the possibility of respondent bias where participants tell the interviewer what they think the researcher wants to hear. Additional methods of data collection, including triangulation, may have strengthened the research design in Phase I.

Response bias may have occurred when respondents are not only affected by their true response to a question, but also how the question is worded or by their own motivations (Gotay, 1996). Response bias in questionnaires or interviews may include yes-saying, end aversion, halo, and framing; these may be prevented through appropriate measure design (Streiner & Norman, 1995; Gotay, 1996). Additional response biases in HRQoL assessment includes social desirability and response shifts, but are reported to be less of a problem in anonymous questionnaires and diaries, and especially in personal narratives and projective techniques (Streiner & Norman, 1995; Gotay, 1996). Parental diaries may have been useful as a complementary method of HRQoL assessment in this research programme by asking parents to describe aspects of their child's HRQoL prospectively; however, diaries require careful training, both initially and on follow-up, to ensure complete and consistent recording (Gotay, 1996).

Financial and time resources limited the systematic review; one person performed the review with a second independent reviewer assessing a subsample of included papers for methodological quality. A review of titles obtained from electronic searches by a second reviewer may have enhanced the reliability of the review.

7.3.1.2 Alternative samples

A control group was not used in the validation phase of this study. Historically, the rigorous standard for assessing potential tradeoffs between the burden of disease and the benefit of therapeutic interventions has been the use of a small 'control sample' in individual investigations. However, such results are often limited because the sample is usually a 'convenience' sample rather than a randomly selected representative group matched to the age and sex distribution of the sample being studied (Landgraf & Abetz, 1998). It is reported that ratings from patients would be easier to interpret if scores for the target sample could be evaluated relative to the reports from a representative population-

based sample or norm (Landgraf & Abetz, 1998). The application of this strategy for assessing HRQoL in children is relatively unexplored, with the exception of the development of the CHQ, a generic HRQoL measure, which was developed specifically for norm-based interpretation of children's physical and emotional functional status and wellbeing (Landgraf & Abetz, 1998). The advantages of normative methods are that individual patients and groups of patients can be compared to population trends, and unusual deviations from the norm can be noted (Jacobson & Fried, 1998).

The PICQoL questionnaire developed in this study is a client-specific HRQoL measure whose purpose is to evaluate the changes in a child's HRQoL when admitted to a PICU with a respiratory illness. The measure is not a generic HRQoL measure, which would allow the comparison of the HRQoL of one child to that of another from a normal healthy population (Eiser & Morse, 2001a). Testing the PICQoL questionnaire in a group of 'non-hospitalised' or 'healthy' children is however recommended as a future area of research to aid interpretation of the PICQoL scores and to test the discriminant validity of the measure.

In evaluating new treatments involving rare conditions, international measures of HRQoL have been advocated (Eiser & Morse, 2001a). A fundamental assumption in assessing HRQoL cross-culturally is the existence of a 'universal' construct, that is, if a particular measure is to be adapted or developed for use in two or more cultures, then it is assumed that HRQoL is an underlying universal construct, assigned the same relevant attributes across cultures (Anderson et al, 1996). This is an assumption of the generality of the construct. A second common assumption is that this universal construct can be appropriately measured by a common set of indicators; this is an assumption of generality of indicators (Anderson et al, 1996). A final common assumption is a generality of scaling in terms of the relationship of the response categories to the underlying dimension measured. Scaling assumptions in cross-cultural context specify what is meant by metric equivalence, that is, whether the meanings of the response categories are the same across cultures or groups (Anderson et al, 1996). Two assessment approaches of HRQoL are reported that can be used cross-culturally: functional-states and personal value judgements (Anderson et al, 1996).

Cross-cultural validation of HRQoL measures should include key psychometric properties of item equivalence, the degree to which the items composing the measure are identical across cultures; scalar equivalence, when a given rating or response is equated to the same

degree of the construct across cultures; and internal structure congruence, when a construct has the same dimensions and the same interrelations among the dimensions across cultures (Anderson et al, 1996). Item equivalence is heavily dependent upon proper translation of the measure; scale equivalence and internal structure congruence pertain to construct validity (Anderson et al, 1996). Operational equivalence, the relative performance of a measure using different modes of administration, is also reported (Anderson et al, 1996). Anderson et al (1996) suggest four methods for examining these assumptions, and each will be discussed in turn.

Firstly multidimensional scaling, where, for example, parents of various cultures rate the degree of similarity or dissimilarity between pairs of HRQoL-related words, phrases or concepts (e.g. 'unable to dress myself' and 'feeling useless'). The HRQoL-related concepts that are judged to be very similar should lie close together in the multidimensional scaling solution.

Secondly, multigroup factor analyses, where, for example, independent random samples of parents from each culture can be obtained and administered identical PICQoL questionnaires. The form of factor structure can then be examined across cultures to see if it is the same, the factor loadings can also be explored for equivalence, and the means of measured variables and latent variables (e.g. subscales such as positive milestones or negative emotions) explored for equivalence, the latter demonstrating scalar equivalence.

Thirdly, hierarchical linear modeling, which investigates construct validity. For example, the PICQoL questionnaire may be known to correlate with a severity of illness variable, the relation between the PICQoL questionnaire and the severity of illness variable is thus measured within cultures and the difference between these relations examined across cultures – the levels of hierarchy may be called within culture or between culture. The goal of the analysis is to determine whether there are significant, statistical differences in the construct validity of the PICQoL questionnaire, represented by the relation between HRQoL and the severity of illness variable, between cultures.

Lastly, differential item functioning (DIF), which determines whether an item is performing differently across cultures. For example, the goal of DIF analysis is to determine whether two parents from different cultural groups with similar levels of the

underlying, latent variable representing the construct HRQoL have the same probability of responding positively to the item. If the two matched parents have different probabilities of answering the question positively, then the item is said to exhibit DIF; if the DIF is large enough then the item is reworded or removed from the PICQoL questionnaire.

Parents who were not English-literate were not sampled in this research programme; this may have led to an under-representation of non-English speaking or reading parents, particularly those from the ethnic minorities. Ten per cent, 17% and 11% of parents who participated in Phases I-III respectively were from the ethnic minorities, which generally under-represents parents admitted to the PICU in this study. Attempts to overcome this under-representation could have been enhanced by over-sampling parents from ethnic minorities and utilising the skills of an interpretator, but the latter consideration was beyond the financial resources available. The parents of children from ethnic minorities sampled in this study did, however, represent the major ethnic groups in the community affected by a respiratory illness.

The sample size and hence response rates could have been enhanced, particularly in Phase III, by recruiting parents from other PICUs. However, this would have impacted upon the costs of the research programme in terms of time and financial resources. The involvement of parents and clinicians from other centres in the item-development and testing stage would also have been necessary to ensure that the measure was representative of the population in which it was developed.

Random sampling methods aimed to ensure that all elements of the population had a chance to be selected in order to obtain a representative sample (Polgar & Thomas, 1998). However, the results of this study can only be generalised to the accessible population under investigation and not the target population of all children under the age of five years admitted to PICU's in the UK with a respiratory illness. Sampling children from other PICU's may strengthen the external validity across the UK.

7.3.1.3 Additional analyses

Greenhalgh and Meadows (1999) describe little evidence to support the effectiveness of incorporating patient-based measures of outcome in routine clinical practice. They state that evidence of the benefit of including these measures in routine practice to clinical

decision-making, resource allocation and health policy is required (Greenhalgh & Meadows, 1999). Clinicians may find information difficult to interpret and perhaps irrelevant (Pham & Klaassen, 2000). The PICQoL questionnaire was field tested in a PICU population but the methods of questionnaire administration and analysis in routine clinical practice were not formally evaluated. The interpretation of PICQoL scores was also not defined, which will be of particular relevance to clinicians and parents. Differing methods of administration of the HRQoL measure are recommended for future research.

Additional statistical analysis may have also enhanced the research design. Performing confirmatory FA on PICQoL and IPQ items may have strengthened the construct validity of the measure (Streiner & Norman, 1995).

7.4 Recommendations for future research

Specific

- The testing of the responsiveness of the HRQoL measure by comparing the mean change in PICQoL scores for a group of stable children and a group of children whose respiratory illness is known to change following a PIC intervention
- Further testing of construct validity using additional constructs such as severity of illness and comorbidity
- Confirmatory FA of PICQoL and IPQ items to test the hypothesised structure of the measure
- A comparison of father/mother PICQoL scores to investigate whether fathers are suitable proxies for the evaluation of HRQoL outcomes
- Investigation of parental anxiety and parental illness perceptions on parental perceptions of HRQoL over time
- Determination of a method to interpret PICQoL scores to assist clinicians in clinical decision-making

General

- The further evaluation of the feasibility or clinical utility of the PICQoL questionnaire in routine clinical practice or clinical research (e.g. parent-completion prior to clinician consultation versus postal parent-completion)

- The testing of appropriate PICQoL items in a healthy child population to determine the discriminant validity of the measure
- A multi-informant assessment of HRQoL using the PICQoL questionnaire, including the views of health care professionals and other professionals, e.g. teachers
- The testing of generic PICQoL items in other patient groups receiving PIC and the development of additional system-specific modules for evaluating HRQoL outcomes following PIC (e.g. neurological-specific items)
- The development of a PICQoL questionnaire manual to explain the development, testing and interpretation of the PICQoL questionnaire
- Evaluation of the PICQoL questionnaire in the current RCT to provide further information on the performance of the measure and to contribute to data interpretation on a range of PICQoL scores
- The further testing of the reliability and validity of Carer-version of the IPQ in a PICU population, including confirmatory FA.
- The active dissemination of the thesis findings to relevant audiences, and development of appropriate implementation strategies, to promote the routine data collection of HRQoL outcomes in children following PIC
- The acquisition of additional research funds to further test the reliability and validity of the PICQoL questionnaire in larger samples, other PIC disease groups, and other PICUs in the UK.

7.5 Conclusion

Because improving health is the ultimate goal of a health care system, the measurement of health outcomes in research is a logical and important goal for the evaluation of the impact of health services (Vivier et al, 1994). Appropriate, rigorously designed and evaluated HRQoL measures can be used in carefully designed studies to provide objective representations of what until recently has been viewed as essentially intangible subjective processes (Schipper et al, 1996).

The evaluation of HRQoL outcomes for children following PIC is a national priority area for research. This thesis has contributed to research in this field by generating new

knowledge. Two important contributions have been made to the evidence base for outcome measurement in PIC.

Firstly, a systematic review and quality evaluation of published child HRQoL outcome measures, and a synthesis of the evidence relating to the development, psychometric properties, user-centredness, clinical utility, and feasibility of outcome measures applied in PIC. Secondly, the development and testing of a system-specific HRQoL measure, the PICQoL questionnaire, for future use in the evaluation of the effectiveness of PIC interventions in the UK through the monitoring of HRQoL outcomes over time from the parent perspective.

Further research is recommended to strengthen the psychometric properties of the HRQoL measure and to address the recognised limitations.

Appendix I

DATA EXTRACTION FORM (Version 2)
HRQoL IN CHILDREN REVIEW*

(Note: The boxes have been reduced to comply with thesis margin requirements and the form printed on single pages)

ID □□□ **Reviewer** [ ] **Date** □□/□□/□□

Name of QoL instrument [ ]

Instrument in file Y/N

Type of instrument
Generic □ Client-specific □
Disease-specific □ Population specific □
Specify disease.....

DETAILS OF PUBLICATION

Author(s) [ ]

Title [ ]

Source and reference [ ]

Institution/contact address [ ]

Country of origin [ ]

RESEARCH QUESTION

Aims of the study

□ Development of QoL measure [ ]

□ Determine statistical properties of QoL measure [ ]

□ Compare proxy ratings [ ]

□ Compare outcomes in clinical trial [ ]

□ Compare treatments [ ]

□ Evaluate intervention [ ]

□ Other (specify) [ ]

DESCRIPTION OF THE MEASURE

Instrument purpose Predictive ☐ Evaluative ☐ Discriminative☐

Rationale stated? Y/N

Pre-existing measure (specify name and changes made)

Did the development and testing of the instrument involve separate stages? Y/N

Is the pilot stage reported? Y/N

Method of item generation (e.g. semi-structured interview)

Existing scales ☐ Child reports ☐ Expert opinion ☐
(who?.....)
Clinical observations ☐ Parent reports ☐ Theory based? ☐
Other ☐
(specify.....)

Design of measure a) Adult ☐ Child-centred ☐ Both ☐

b) Quality of life ☐
Health status ☐
Functional status ☐
Preference/utility based ☐
Other ☐
(specify.....)

Definition of QoL Multidimensional/WHO ☐ Cost-effectiveness ☐
Goal-orientated ☐

Has the measure taken account of sensitivity to developmental milestones? Y/N
Has the measure considered the impact of cognitive develop. on evaluations of HRQoL? Y/N

Instrument completed by? Parent ☐ Clinician ☐
Child ☐ Other [ ]

Age limit [ ] years

Number & name of domains [ ]

Total number of items [ ]

Proxy ratings available? Y/N

Response format E.g. visual analogue, Likert scale

[ ]

Did questions involve assessment across time frames? (E.g. last month, last week) Y/N

Instrument scoring How has the HRQoL score been classified?
☐ Single indicator ☐ Profile ☐ Battery

Normative data available? Y/N

PROCEDURE (indicate 'not reported' if information is not given)

STUDY DESIGN

Design of study

RCT ☐
Cohort study with matched concurrent controls ☐
Cohort study with unmatched concurrent controls ☐
Cohort study with historic controls ☐
Cohort study with no controls ☐
Other (specify).....

Method of randomisation [ ]

Inclusion criteria [ ]

Exclusion criteria [ ]

Recruitment procedures [ ]

SAMPLE CHARACTERISTICS

Scale of the study Number of participants in each condition/target group/control

Number of participants in the final analysis

Population characteristics Mean age Age-range

Gender % male %female

Ethnicity

Age at diagnosis

Time since diagnosis

Time since completion of treatment

Representativeness of the sample

Number of participants v. non-participants

Informant (e.g. child, mother, father, clinician, teacher)

Method of administration and by whom (e.g. post/phone/interview)

Length of follow-up (complete if longitudinal design)

EVALUATION

DEVELOPMENT OF AN INSTRUMENT

Validation measures used

Child completed

Parent completed

Clinical

CROSS-SECTIONAL STUDIES

Baseline measures (specify whether parent or child completed)

LONGITUDINAL STUDIES

Follow-up measures

Statistical techniques used

Attrition rate

How was attrition managed?

Number followed up in each condition

RESULTS

Quantitative results (Report correlation coefficients where necessary. Specify differences between different populations, and differences between domains of QoL)

Reliability Internal consistency

Test-retest reliability

Inter-rater reliability

Validity Face validity

Content validity

Criterion validity

Construct validity (convergent/ discriminative)

Responsiveness

Responsiveness to change

External variables used? Y/N
Interventions of known efficacy given? Y/N

Practicality

Any training needed to administer instrument? Y/N

Completion time ..... mins.

Feasibility

Feasible for application area? Y/N

Data quality

Ease of analysis and interpretation of scores

Feedback to child/parent or clinician

Likelihood of bias

Comprehensiveness

User-centredness

Does the measure capture the desired outcomes of the parent/child or clinician? Y/N
Does it provide an insight into health care user's views? Y/N

Is the measure faithful to the content of health care user's, carer and/or clinician views?	Y/N
Does the method of obtaining, coding or analysing views distort them?	Y/N
Does any standardisation distort views?	Y/N
Were items checked for high/low endorsement frequency, restriction in range of answers, comprehensibility and ambiguity of phrases or possible offensive content?	Y/N

Utility

Is the instrument acceptable to users/carers or clinicians?	Y/N
Does it provide extra information not already available?	Y/N
Can the measure become an integral part of data collection and thus aid treatment and decision making?	Y/N

QUALITY OF THE STUDY

Effectiveness of measures (author's conclusions)

Effectiveness of measures (reviewer's conclusions if different from above)

Limitations of measure and procedure (author's comments)

Limitations of measure and procedure (reviewer's comments if different from above)

Measure included with the article Y/N

Quality Score:

Reliability	0	1	2	**Total score:**
Validity	0	1	2	
Responsiveness	0	1	2	
Practicality	0	1	2	Note:
Feasibility	0	1	2	0 = failed or did not mention criterion
User-centredness	0	1	2	1 = criterion partially fulfilled
Utility	0	1	2	2 = prima facie evidence that criterion met

REVIEWER'S DECISION

Does the study address the following?

Validity of the measure Y/N

Reliability of the measure Y/N

Is the paper to be included? Yes ☐ No ☐ Unsure ☐

*Adapted from a data extraction form described by Eiser & Morse (2001a) and a critical appraisal checklist for reviewing outcome measures in routine practice by Greenhalgh et al (1998).

References

Eiser C & Morse R (2001a) Quality-of-life-measures in chronic diseases of childhood. *Health Technology Assessment,* 5 (4).

Greenhalgh J, Long A, Brettle AJ & Grant MJ (1998) Reviewing and selecting outcome measures for use in routine practice. *Journal of Evaluation in Clinical Practice,* 4 (4): 339-350.

Appendix II Summary of excluded generic HRQoL measures

Measure	Study	Origin	Age-limit (years)	Respondent	No. of items (dimensions)	Name of dimensions	Psychometric properties			
							Reliability	Validity	Responsiveness	Practicality
Adolesc-ent & Child Health Illness Profile	Starfield et al (1993 & 1995); Riley et al (1998)	USA	11-17	Adolescent	153 (6)	Achievement, comfort, disorders, resilience, risks & satisfaction	Good reliability & internal consistency (>0.7), good test-retest reliability	Good construct, convergent & divergent validity	Not tested	Fair - 30 minutes
Dartmouth COOP charts	Nelson et al (1987; 1990 & 1996)	USA	8-18	Adolescent & child, or proxy	6 (6)	Physical fitness, emotional feelings, schoolwork, social support, family communication & health habits	Good test-retest reliability for adolescent charts (0.77). Intraclass correlation 0.77. Child charts less reliable	Good validity across a range of socio-economic and health groups	Uncertain	Excellent clarity to users and use in clinical practice, few minutes to complete
15D, 16D & 17D HRQOL	Apajasalo et al (1996a & 1996b; 1997 & 1998)	Finland	12-15 (16D) 8-11 (17D)	Adolescent Child	16 D: 16 (16) 17D: 17 (17)	16D – mobility, vision, hearing, breathing, sleeping, eating, elimination, speech, mental function, discomfort & symptoms, school & hobbies, friends, physical appearance, depression, distress & vitality. 17D - as for 16 D plus anxiety, ability to concentrate, learning ability & memory	Internal consistency is unclear. Test-retest reliability was recorded. Assumptions made about reliability & validity as adapted from 15D.	Good criterion & construct validity	Some evidence of differentiating HRQoL of patient groups and healthy children.	Easy to use, 10-15 minutes
HAY	Bruill et al (1997); Le Coq et al (2000a, b & c)	The Nether-lands	8-13	Parent & child	80 (5 generic; 4 asthma)	Physical, cognitive & social functioning, physical complaints & happiness	Internal consistency is reported as 0.71-0.83. Test-retest reliability not described.	Validity yet to be assessed	Responsiveness yet to be assessed	Not reported

Appendix II Summary of excluded generic HRQoL measures

Measure	Study	Origin	Age-limit (years)	Respondent	No. of items (dimensions)	Name of dimensions	Psychometric properties			
							Reliability	Validity	Responsiveness	Practicality
TACQOL	Theunissen et al (1998); Vogels et al (1998); Verrips et al (1997 & 1999)	The Nether-lands	6-15 parent form 8-15 child form	Parent & child (different forms)	108 (7) - parent form ? child form	Pain & symptoms; basic motor, cognitive & social functioning; global positive & global negative emotional functioning.	Internal consistency reliability (parent form) 0.71-0.89, and 0.59-0.86 (child form). Test-retest reliability not checked.	Face validity good. Convergent validity is poor. Criterion validity tested with KINDL measure, Pearson correlations 0.24-0.60	Not tested	Good - 10 minutes
EHRQL	Eiser et al (1995 & 1999); Eiser, Mohay & Morse (2000); Eiser, Vance & Seamark (2000)	UK	6-11	Adolescent	16 items (0)	Not reported	Moderate internal reliability (0.56-0.64). Test-retest reliability not checked	Satisfactory criterion validity. Construct validity tested between asthmatic and healthy children – satisfactory	Not tested	Interview administered, but child complete – 20 min. Time intensive but enjoyable for children
CQOL	Graham, Stevenson & Flynn (1997); Titman et al (1997)	UK	9-15	Child & parent	15 items (15)	Getting about & using hands, school, out-of-school activities, friends, family relationships, discomfort due to bodily symptoms, worries, depression, seeing, communication, eating, sleep & appearance	Internal consistency reported as satisfactory (0.81-0.87). Test-retest reliability is poor	Face validity and criterion validity reported as satisfactory	Limited evidence	Good - 10-15 minutes for both versions
GQL	Collier & MacKinlay (1997); Collier, MacKinlay & Phillips (2000)	UK	6-16	Child & parent	25 items (5)	General effect, peer relationships, attainments, relationships with parents & general satisfaction	Internal consistency & test-retest reliability not tested.	Content validity is satisfactory, face validity is weak. Criterion & construct validity - not tested. Satisfactory discriminant validity - briefly assessed.	Not tested	Not tested, scale is reported as child-friendly and children understood the scale.

Appendix II Summary of excluded generic HRQoL measures

Measure	Study	Origin	Age-limit (years)	Respondent	No. of items (dimensions)	Name of dimensions	Psychometric properties			
							Reliability	Validity	Responsiveness	Practicality
KINDL	Ravens-Sieberer & Bullinger (1998)	Germany	8-16	Child & parent	40 items (4)	Psychological well-being, social relationships, physical functioning & everyday life activities	Internal consistency - good (>0.75 for subscales, 0.95 for total scale); test-retest reliability stated - results not reported	Face validity good – items developed from child interviews. Criterion validity high, discriminant validity poor. Construct validity not reported	Not assessed	Good – 12 minutes for child version. Children understood scale.
QLQC	Bouman et al (1999)	The Nether-lands	8-12	Child & parent	118 items (3)	Physical, psychological & social functioning	Internal reliability - good (above 0.80 for domain scales). Test-retest reliability - good for psychological & social domains; weak for physical domain.	Convergent validity - - good for aspects of functioning. Discriminative validity insufficient in detecting real HRQoL differences. Criterion validity assessed.	QLQC is reported as sensitive, but not tested over time (other than for test-retest reliability check)	Good – 15mins for child complete version. No comment on parent-complete version
C-QOL	Jirojanakul & Skevington (2000)	Thailand	5-8	Child & parent	Initially 62, reduced to 54 in both forms (? 6)	Physical, psychological, levels of independence, social relationships, environment, spirituality, religion & personal beliefs; 2 global QoL items.	Good internal consistency reliability for child form 0.86 and 0.84 for the parent form. Good test-retest reliability: 0.91 for child form & 0.90 for parent form.	Adequate face and content validity. Discriminant validity checked but requires further investigation on a larger sample. Criterion/construct validity not assessed.	Not tested.	? interview administered Child complete: 20-30 min – fair. Completion time on carer form not reported.
Quality of Life Profile – Adolesc-ent Version: QOLPAV	Raphael et al (1996)	Canada	14-20	Adolescent	54 items (3) (9 sub-domains)	Being (physical, psychological, spiritual); belonging (physical, social, community); becoming (practical, leisure growth)	Good internal consistency (0.94 for all items)	Criterion validity tested against self-esteem, life satisfaction, social support & life chances scales – sig. correlations (0.24-0.56). Construct validity was poor.	Not reported	Lengthy – 40 min.

Appendix II Summary of excluded generic HRQoL measures

Measure	Study	Origin	Age-limit (years)	Respondent	No. of items (dimensions)	Name of dimensions	Psychometric properties			
							Reliability	Validity	Responsiveness	Practicality
VSP-A	Simeoni et al (2000)	France	11-17	Adolescent	40 (6)	Psychological well-being, energy, friends, parents, leisure & school	Good internal consistency (0.83) and test-retest reliability (r=0.69)	Content and construct validity were confirmed	Not reported	<15 minutes
CHQ	Landgraf & Abetz (1996 & 1997); Landgraf et al (1996 & 1998); Epker & Maddray (1998); McGrath et al (1999); Waters et al (1999 & 2000); Asmussen et al (2000); Landgraf (2000); Waters, Salmon & Wake (2000)	USA	5-15	Parent & child (10-15 yr.)	Parent –28, 50, 98 & 135 Child – 87 (14, except 12 in CHQ-PF50)	Physical functioning, role/social functioning (physical), general health perceptions, bodily pain, discomfort, general behaviour, mental health, self-esteem, role/social functioning (emotional), parental impact (emotional), parental impact (time), family activities & family cohesion; global item – change in health.	Good internal consistency (0.62-0.91) & reliability of CHQ-CF87	Good discriminant validity & construct validity	CHQ-PF98 is responsive to children varying in hyperactivity disorder & other clinical conditions	Short form available
ComQol	Gullone & Cummins (1999)	Australia	12-18	Adolescent	? (7)	?	Internal consistency analysis and test-retest reliability reported as adequate	Convergent validity tested	Not reported	Not reported

Appendix II Summary of excluded generic HRQoL measures

Measure	Study	Origin	Age-limit (years)	Respondent	No. of items (dimensions)	Name of dimensions	Psychometric properties			
							Reliability	Validity	Responsiveness	Practicality
MMQL	Not published	US	8-12 13-20	Adolescent	8-12 yrs: 47 (?) 13-20 yrs: 98 (?)	?	?	?	?	Telephone interview

Appendix II Summary of excluded respiratory-specific HRQoL measures

Measure	Study	Origin	Age-limit (years)	Respondent	No. of items (dimensions)	Name of dimensions	Psychometric properties			
							Reliability	Validity	Responsiveness	Practicality
Pediatric Asthma Quality of Life Questionnaire (PAQLQ)	Gibson et al (1995); Juniper et al (1996); Elizabeth et al (1999); Reichenberg & Broberg (2000); Juniper et al (1996 & 1997)*; Clarke et al (1999)	Canada	7-17	Child	23 (3)	Activity limitation, symptoms, & emotional function	Test-retest reliability - intra-class correlation 0.95 for stable patients	Construct (concurrent) validity - significant correlations between PAQLQ & clinical measures (FEV1, PEFR, B-agonist use) & with feeling thermometers	Tested by assessing within-subject changes for those children that changed between the 2 study periods	10-15 minutes
About my Asthma (AMA)	Mishoe et al (1998)	USA	6-12	Child	44 (none)	Unitary scale not grouped into dimensions	Test-retest reliability: α=0.93	Construct (concurrent) validity reported - moderate	Not reported	15-20 mins.
Integrated Therapeutics Group Child Asthma Short Form	Bukstein et al (2000)	USA	?	Parent	17 reduced to 8 (3)	Day-time & night-time symptom scales; and functional limitations scale	Good internal consistency α>0.7	Clinical validity supported	Not reported	Good - brief

Note: *Also used the HUI measure.

Appendix III – Parental invitation to interview, information sheet and consent form (Phase I)

Ward 2, Children's General Intensive Care
C Floor, Jubilee Wing, Leeds General Infirmary
Great George Street, Leeds LS1 3EX

Enquiries to: Angela Grange

Direct Line: 0113 392 3220

Date: (today's date)

Dear (name of parent),

My name is Angela Grange and I am a children's nurse with experience in children's intensive care nursing. I am currently working full-time on Ward 2 (children's general intensive care unit - PICU), Leeds General Infirmary, on a large research project funded by the NHS. Recently Dr Mark Darowski and Michelle Milner wrote to you about this project and explained that I would be contacting you soon to invite you to take part in the project. The project aims to look at how a child's health and lifestyle is affected following a stay on the children's intensive care unit.

I would like to invite you and/or your partner to take part in this research project. I wondered whether you and/or your partner would be willing to be interviewed by me for about an hour. I would like to discuss (name of child) health and wellbeing since discharge from the children's intensive care unit a few months ago. This informal interview can either take place in your own home or in the hospital, Leeds General Infirmary. I will refund any travelling expenses for a hospital-based interview. Please be reassured that any information you give to me will be treated confidentially and anonymously. Please find enclosed a parental/carer information sheet explaining the research project in more detail.

If you agree to take part, I would be most grateful if you could complete the attached form and return it to me in the stamped addressed envelope provided. This will help me to arrange a suitable time and place for the interview at your convenience. I will contact you soon by telephone/letter to arrange an appropriate date and time.

If you have any questions about this project, please do not hesitate to contact me on the telephone number/address above. I look forward to hearing from you and thank you in anticipation for your support in this project.

Yours sincerely,

Mrs Angela Grange
Research Fellow/Registered Sick Children's Nurse

CHILDREN'S INTENSIVE CARE RESEARCH PROJECT

Please complete in block capitals.

NAME:
(Mr/Ms/Mrs/Miss/Dr) .....

ADDRESS:
(Street) .....

(Area) .....

(City) .....

(Postcode) .....

TELEPHONE:
(Daytime) .....

(Evening) .....

(Please indicate a suitable date and time for an interview overleaf)

P.T.O.

Please indicate in the table below the most convenient date and time for you and/or your partner to take part in an interview (please tick as many boxes that apply):

Date	Morning	Afternoon	Evening
June 1st (Tues)			
June 2nd (Wed)			
June 3rd (Thurs)			
June 4th (Fri)			
June 5th (Sat)			
June 6th (Sun)			
June 7th (Mon)			
June 8th (Tues)			
June 9th (Wed)			
June 10th (Thurs)			
June 11th (Fri)			
June 12th(Sat)			
June 13th (Sun)			
June 14th (Mon)			
June 15th (Tues)			
June 16th (Wed)			
June 17th (Thurs)			
June 18th (Fri)			
June 19th (Sat)			
June 20th (Sun)			

If none of the above dates are convenient, please indicate a date and time that would be more suitable:

Date: ------/------/------ **Time:** **am/pm**

Would you prefer the interview to take place in your home or at the hospital (Leeds General Infirmary)? (*Please tick the appropriate box below)*

Home ☐ Hospital ☐

Any questions or comments?

Thank you for your support.
Please return to Angela Grange in the envelope provided.

Appendix III (continued)

Parent/Carer Information Sheet

Children's Health and Wellbeing after Intensive Care

Dear parent/carer,

I would like to invite you to take part in a research project.

My name is Angela Grange and I am a qualified children's nurse specialising in children's intensive care nursing. I am currently working as a research fellow on Ward 2 (children's general intensive care unit/PICU) at the Leeds General Infirmary. My post and research project are funded by a grant from the NHS.

In this project I would like to look at how a child's health and wellbeing are affected following their discharge from the children's intensive care unit. I would like to do this from the viewpoint of the child's main carer/parent. The information that carers/parents give me will help me to develop a short questionnaire. This questionnaire will be used in the future to measure a child's health and wellbeing following their discharge from the children's intensive care unit.

I hope that the information from this project will improve the delivery and quality of care that children and parents receive on the children's intensive care unit. Additionally, I hope that this project will give staff on the children's intensive care unit a clearer picture of the impact of their care on the child's health and wellbeing following their discharge. This will help the staff to monitor and evaluate the care and services that they provide.

If you decide to take part I would like to ask you a few questions about your child's health and wellbeing following their discharge from the children's intensive care unit. This should take about an hour and, if it is convenient, I would like to visit you in your home to ask these questions.

Taking part in this project will not benefit you or your child and is entirely voluntary. If you do not wish to take part, or you wish to withdraw at any time, you are completely free to do so. If you agree to take part, any information that you give to me will be treated confidentially and anonymously, and will be destroyed at the end of the study.

If you have any questions, please do not hesitate to contact me. My contact details are shown below.

NAME	Mrs Angela Grange
ADDRESS **Work:** **Home:**	 C/o Ward 2 Children's General Intensive Care Unit C Floor Jubilee Wing Leeds General Infirmary Great George Street Leeds Teaching Hospitals LS1 3EX 13 Granville Terrace Otley Leeds LS21 3EJ
TELEPHONE **Work:** **Home:** **Mobile:**	 0113 392 3220/7102 01943 462 159 0585 306 310

Appendix III (continued)

*Note: Original in size 14 font

Parent/Significant Carer Consent Form*

I, _____ (your name) give Angela Grange (Research Fellow, Ward 2) permission to interview me in my own home/hospital for the research project *"Children's Health and Wellbeing after Intensive Care".*

I also agree to Angela Grange obtaining information about my child's care from other resources, e.g. hospital doctor, general practitioner, health visitor, schoolteacher/nursery teacher, psychologist, other members of the health care team, and patient records.

I understand the purpose of the project and realise that I can withdraw my consent at anytime without detriment to my child or myself.

Signature _____

Name (Please print) _____

Address for correspondence_____
(Please print)

_____

_____

_____

_____ (Postcode)

Telephone _____

Relationship to child _____

Date _____

Please indicate if you would like a summary of the research results:

Yes ☐ No ☐

I, Angela Grange, confirm that I have explained to_____
(Parent/carer's name) the purpose of the research project in a way they understand.

Signature _____

Date _____

Appendix IV – Definition of themes and categories (Phase I)

NUD*IST CODE	THEME (DEFINITION)	CATEGORIES	DEFINITION OF CATEGORY
2	**MEDICAL HISTORY (Parent's perception of their child's medical history)**	1 Age of child	1 Age of child on admission to PICU
		2 Length of PICU stay	2 Child's PICU length of stay
		3 Previous hospital admissions	3 Number of child's hospital admissions (prior to interview date)
		4 Previous PICU admissions	4 Number of child's PICU admissions (prior to interview date)
		5 LOS at DGH pre PICU	5 Child's length of stay at the referring hospital prior to PICU admission
		6 LOS at DGH post PICU	6 Child's length of stay on the ward (DGH or LGI) post PICU discharge
		7 Total length of hospital stay	7 Child's total length of hospital stay (including pre and post PICU care)
		8 Maternity history	8 Maternity history, e.g. mode of delivery, gestation, birth order, birth weight
		9 Pathway of care to PICU	9 Care pathway to PICU, e.g. admitted to local hospital, consultant/GP called, etc.
		10 Pathway of care from PICU	10 Care pathway from PICU, e.g. transferred back to DGH, other referrals, etc.
		11 Reason for previous/subsequent admission	11Reason for child's other hospital admissions, e.g. surgery, asthma, etc.
		12 Time at home out of hospital	12 Amount of time spent at home by child with chronic illness e.g. week, few days, etc.
		13 Place of previous hospital admission	13 Name of ward or hospital where other hospital admissions took place.
3	**STRESSORS - FACTORS CAUSING PARENTS STRESS DURING THEIR CHILD'S ILLNESS (Factors that parents perceived to cause them stress during their child's illness – pre, during and post PICU)**	1 Child's condition	1 Child's condition, e.g. severity of illness, vulnerability, no change, seeing child upset, etc.
		2 Ward environment	2 Ward environment (at DGH or LGI), e.g. equipment, surroundings, etc.
		3 PICU environment	3 PICU environment, e.g. equipment, routine, drugs, condition of other children, etc.
		4 Level of health care delivered	4 Standard of health care delivered in PICU/wards, e.g. limited experience of staff, inconsistency, attitude of health care staff, differences between DGH and PICU etc.
		5 Health care professionals communication	5 Communication by health care staff, e.g. to parents, between nurses, information not openly shared with parents, etc.
		6 Partner relationship	6 Partner relationships, e.g. strain, communication breakdown, rows with partner, etc.
		7 Family relationships	7 Family relationships, e.g. anxiety of other family members, visiting, etc.
		8 Parental role	8 Loss of parental role, e.g. shift in responsibility, lack of specialist skills, etc.
		9 Stressors apart from child's illness crisis (external) - none, travel, work	9 Other stressors, e.g. work commitments, travelling to hospital, financial strain, etc.
		10 Managing child at home prior to PICU episode	10 Caring for child at home pre PICU admission, e.g. treatment ineffective at home, child cried all the time, parent felt unprepared to deal with resuscitation, etc.
		11 Managing child at home post PICU episode	11 Caring for child at home post PICU discharge, e.g. lack of monitoring, managing drugs, planning trips out, assuming total responsibility for child, etc.

Appendix IV (continued) – Definition of themes and categories (Phase I)			
NUD*IST CODE	**THEME (DEFINITION)**	**CATEGORIES**	**DEFINITION OF CATEGORY**
4	**PARENTAL WELLBEING (Parental perception of their own general wellbeing during their child's illness crisis)**	1 Expression of emotions 2 Feelings of hope 3 Physical signs and symptoms (parent)	1 Parental expressions of emotion, e.g. fear – such as fear of future admissions/illness, anxiety (upset), shock, denial, anger, disbelief, etc. 2 Parental feelings of hope, e.g. for recovery, child is stable, not hopeful, etc. 3 Physical signs and symptoms exhibited by parents, e.g. ↑heart rate, sleeplessness
5	**PARENTAL UNDERSTANDING OF CHILD'S ILLNESS (Parental perception of their child's own illness – based upon Leventhal's model of illness representations, including identity, cause, timeline, cure and control. Note: consequences are dealt with in themes 18-22)**	1 Social advice (PICU illness) 2 Family advice (PICU illness) 3 Health care professional advice (PICU illness) 4 Advice on additional illness (es) 5 Respiratory symptoms of PICU illness 6 Other symptoms of PICU illness (non-respiratory) 7 Symptoms of additional illnesses 8 Diagnosis of PICU illness 9 Diagnosis of additional illness (es) 10 Cause of PICU illness 11 Cause of additional illness (es) 12 Duration of symptoms of PICU illness 13 Duration of symptoms of additional illness (es) 14 Cure/control of PICU illness 15 Cure/control of additional illness (es) 16 Time for child to return to normal 17 Time for parent to return to normal	1 Social advice on child's illness, e.g. friends and neighbours 2 Advice on child's illness from family members, e.g. grandparents, etc. 3 Advice on child's illness from health care staff, e.g. GP, HV, midwife, nurse, etc. 4 Advice given on additional illness of child, e.g. social, family and health care 5 Respiratory signs and symptoms of child's PICU illness (note: cyanosis=going blue) 6 Other symptoms of child's PICU illness that are not respiratory related, e.g. vomiting, temperature, loss of appetite, lifeless, etc. 7 Child's symptoms of additional illnesses, e.g. swelling on neck, fever, cough, etc. 8 Diagnosis of the child's PICU illness, e.g. bronchiolitis, asthma etc. 9 Diagnosis of the child's additional illnesses, e.g. neutropenia, reflux, etc. 10 Cause of child's PICU illness, e.g. virus, infection from birth, immune suppressed, 11 Cause of child's additional illnesses, e.g. side effect of immunisation, born too quickly, caught from siblings, etc. 12 Length of time of onset/duration of symptoms of PICU illness, e.g. few days, etc. 13 Length of time of onset/duration of symptoms of additional illness 14 Cure or control of child's PICU illness, e.g. uncertain outcome, improves with nebulisers, etc. 15 Cure or control of child's additional illnesses, e.g. requires further surgery, 16 Length of time taken for child to return to normal routine, health, e.g. a week, etc. 17 Length of time taken for parent to return to normal way of life, e.g. few days, etc.

Appendix IV (continued) – Definition of themes and categories (Phase I)			
NUD*IST CODE	**THEME (DEFINITION)**	**CATEGORIES**	**DEFINITION OF CATEGORY**
6	**COPING WITH THE CHILD'S ILLNESS (Coping mechanisms used by parents during their child's illness crisis)**	1 Parents seeking information and support 2 Family support (visiting) 3 Information sharing from health care professionals 4 Coming to terms with the situation 5 Other 6 Attitude of health care staff	1 Parents seeking information and support, e.g. asking nurse questions, telephone support line, etc. 2 Family support, e.g. parents resident, family visited, looking after siblings, etc. 3 Info. given by health care staff, e.g. explanation of care/equipment & reassurance 4 Methods by which parents come to terms with situation, e.g. accepting outcome, positive coping, etc. 5 Other coping mechanisms, e.g. developing daily routine, support from other parents 6 Attitude of health care staff, e.g. nice staff, approachable, supportive, etc.
7	**GLOBAL GOOD QUALITY OF LIFE (Parental perception of a child who has a good quality of life in global terms, not specifically their own child or siblings)**	1 Emotion 2 Behaviour 3 Cognition 4 Family 5 Environment 6 Inter-personal interaction 7 Health 8 Nutrition	1 Emotional characteristics of the child, e.g. happiness, has attention, loved, laughs 2 Behavioural characteristics of the child, e.g. playful, avoids crime, etc. 3 Cognition characteristics of the child, e.g. good qualifications, school, reads, etc. 4 Family characteristics e.g. brought up by parents, being in a family, etc. 5 Environmental characteristics, e.g. child is clean, warm, lives in nice area, etc. 6 Inter-personal interaction with the child, e.g. socialises with children/adults, etc. 7 Child's general level of health, e.g. no illness, health is monitored regularly, etc. 8 Nutritional characteristics of child, e.g. child is well fed, drinks well, etc.
8	**SPECIFIC GOOD QUALITY OF LIFE (Parent's perception of a child with a good quality of life with reference to their own child or siblings)**	1 Inter-personal interaction 2 Behaviour 3 Family 4 Emotion 5 Own child's quality of life (parental perception) 6 Diet	1 Inter-personal interaction of own child, e.g. independence, parent talks to child & pays attention to child, etc. 2 Behavioural aspects of own child, e.g. playful, child is not spoilt, etc. 3 Family aspects of own child, e.g. parents care for child / spend time with child 4 Emotional aspects of own child, e.g. child is happy, laughs, etc. 5 Parental perception of own child's quality of life, e.g. positive, could be better, etc. 6 Dietary aspects of own child, e.g. eats well, etc.
9	**GLOBAL POOR QUALITY OF LIFE (Parent's perception of a child who has a poor quality of life in global terms)**	1 Emotion 2 Environment 3 Family 4 Health 5 Inter-personal interaction 6 Diet	1 Emotional characteristics of the child, e.g. unhappy, no love, neglected, etc. 2 Environmental aspects, e.g. unsafe, no toys, etc. 3 Family aspects, e.g. parents disinterested in child, no time for play, etc. 4 Health aspects, e.g. poor social background affects health, causes of ill health. 5 Inter-personal interaction with child, e.g. child is not allowed to mix, child is unable to integrate, child is left alone, etc. 6 Dietary aspects, e.g. poor diet, underweight, etc.

Appendix IV (continued) – Definition of themes and categories (Phase I)			
NUD*IST CODE	**THEME (DEFINITION)**	**CATEGORIES**	**DEFINITION OF CATEGORY**
10	**SPECIFIC POOR QUALITY OF LIFE (Parent's perception of a child with a poor quality of life with reference to their own child or siblings)**	1 Inter-personal interaction 2 Behaviour	1 Inter-personal interaction of own child, e.g. child is unable to talk to others, parents disinterested, etc. 2 Behavioural aspects of own child, e.g. tantrums.
11	**DISABILITIES AND QUALITY OF LIFE (Parental perceptions of the quality of life of children with disabilities, and factors that influence these perceptions)**	1 Factors affecting parental perception of quality of life of a disabled child 2 Parental assessment of quality of life in disabled children	1 Factors affecting QoL perceptions of children with disabilities, e.g. level of severity, type of care needed, child's age, experience of disabled children, etc. 2 Parental assessment of QoL of a disabled child, e.g. good, poor, difficult, etc.
12	**FACTORS AFFECTING PARENTAL QUALITY OF LIFE PERCEPTIONS (Factors that parents described that influence their perception of QoL)**	1 Experience with children 2 Parental beliefs and values 3 Parental understanding of the quality of life concept	1 Parent's experience of children, e.g. limited, none, comparison with families, etc. 2 Parent's own beliefs and values, e.g. what is best for child, money isn't important, time with child is important, views on emotional, environmental and social aspects etc. 3 Parent's understanding of the QoL concept, e.g. difficult to explain good QoL, difficult to identify most important aspect, etc.
13	**GLOBAL HEALTH (Parental perceptions of a child who has good health in global terms, i.e. not specifically their own child or siblings)**	1 Emotion 2 Behaviour 3 Cognition 4 Inter-personal interaction 5 Growth and development 6 Diet 7 Environment 8 Family 9 Appearance	1 Emotional aspects of a healthy child, e.g. happy, loved, smiling, etc. 2 Behavioural aspects of a healthy child, e.g. mischievous, playing, no tantrums, etc. 3 Cognitive aspects of a healthy child, e.g. normal mental development, alert, etc. 4 Inter-personal interaction of a healthy child, e.g. actively involved, mixes with friends 5 Growth and development aspects of a healthy child, e.g. mobility, normal progress, speech and communication, etc. 6 Dietary aspects of a healthy child, e.g. well fed, drinks well, etc. 7 Environmental aspects of a healthy child, e.g. clean home, clothes, warm, etc. 8 Family aspects of a healthy child, e.g. parents look after child, child has a home, etc. 9 Appearance of a healthy child, e.g. complexion, looks normal, colour okay, etc.

Appendix IV (continued) – Definition of themes and categories (Phase I)			
NUD*IST CODE	**THEME (DEFINITION)**	**CATEGORIES**	**DEFINITION OF CATEGORY**
14	**SPECIFIC HEALTH (Parental perceptions of a healthy child with reference to their own child or siblings)**	1Emotion 2 Inter-personal interaction 3 Diet 4 Growth and development 5 Own child's health state pre PICU (parental perception) 6 Own child's health state post PICU (parental perception) 7 Behaviour 8 Appearance	1 Emotional aspects of own child, e.g. content, happy, etc. 2 Inter-personal interaction of own child, e.g. with peers, playing, etc. 3 Dietary aspects of own child, e.g. eats a balanced diet, etc. 4 Growth and development aspects of own child, e.g. normal development for age, talking, mobile, etc. 5 Parent's perception of their own child's health state pre PICU, e.g. fine, not healthy, unhealthy, healthy apart from chest, etc. 6 Parent's perception of their own child's health state post PICU, e.g. worse, the same, still has a bad chest, regression in development, etc. 7 Behavioural aspects of own child, e.g. plays, determined, more energy, etc. 8 Appearance of own child, e.g. looks normal, not pale, etc.
15	**GLOBAL UNHEALTHY (Parental perception of a child who has poor health in global terms, i.e. not specifically their own child or siblings)**	1 Emotion 2 Environment 3 Behaviour 4 Health 5 Diet 6 Inter-personal interaction 7 Appearance	1 Emotional aspects of an unhealthy child, e.g. cries, unhappy, etc. 2 Environmental aspects of an unhealthy child, e.g. smokey home, unclean, etc. 3 Behavioural aspects of an unhealthy child, e.g. moody, temperamental/introvert 4 Health aspects of an unhealthy child, e.g. no GP visits, poorly, develop. delay 5 Dietary aspects of an unhealthy child, e.g. loss of appetite, doesn't eat, etc. 6 Inter-personal interaction of an unhealthy child, e.g. child doesn't interact, no attention 7 Appearance of an unhealthy child, e.g. pale, drawn, etc.
16	**SPECIFIC UNHEALTHY (Parental perception of an unhealthy child with reference to their own child or siblings)**	1 Emotion 2 Behaviour 3 Symptoms of illness/disease characteristics 4 Diet 5 Growth and development 6 Treatment	1 Emotional aspects of own child, e.g. sad, unhappy, etc. 2 Behavioural aspects of own child, e.g. disinterested, tantrums, quiet, etc. 3 Symptoms of illness of own child, e.g. temperature, cold, pain, etc. 4 Dietary aspects of own child, e.g. eats crisps and biscuits, doesn't eat, etc. 5 Growth and development aspects of own child, e.g. small for age, regression. 6 Treatment of own child, e.g. requires nebulisers, oxygen-dependent, etc.
17	**FACTORS AFFECTING PARENTAL CHILD HEALTH PERCEPTIONS (Factors that influence parental perceptions of a child's health)**	1 Experience of healthy children 2 Parent's values and beliefs 3 Parental intuition 4 Experience of ill children 5 Parental understanding of the concept of health and ill-health 6 Parent attitudes to health	1 Experience of healthy children, e.g. other siblings' health state, amount of time spent with the child relative to its age, parent's experience of children, etc. 2 Parent's own beliefs and values, e.g. parent's own upbringing, happiness is most important thing, cannot tell a healthy child by looking at it, etc. 3 Parental intuition, e.g. knew something was wrong/child unwell, gut feeling, etc. 4 Parental experience of ill children, e.g. knowledge of hospitals, little experience 5 Parental understanding of health and ill health, e.g. absence of disease, difficult to describe, emotional and physical aspects, etc. 6 Parent's attitudes to health, e.g. disability is a strain, good health=good QoL.

Appendix IV (continued) – Definition of themes and categories (Phase I)			
NUD*IST CODE	**THEME (DEFINITION)**	**CATEGORIES**	**DEFINITION OF CATEGORY**
18	**POSITIVE CONSEQUENCES OF CHILD'S ILLNESS (Parental perception of the positive changes in their own child's health and wellbeing post PICU discharge)**	1 Diet 2 Emotion 3 Health 4 Behaviour 5 Cognition 6 Parent's relationship and wellbeing 7 Respiratory 8 Development 9 Sleep	1 Dietary aspects, e.g. eats more, gaining weight, drinks well, etc. 2 Emotional aspects, e.g. closer bond with mother, not emotionally harmed, happy, 3 Health aspects, e.g. PICU care not harmed child, mostly positive, sees GP quicker, 4 Behavioural aspects, e.g. more strong willed, determined, etc. 5 Cognitive aspects, e.g. more alert, recognises faces, etc. 6 Parent's relationship and their wellbeing, e.g. mother more confident, more knowledgeable of illness, etc. 7 Respiratory symptoms, e.g. improvement in breathing, stopped wheezing, ↓ O_2 8 Developmental aspects, e.g. hearing okay, more mobile, talks more, etc. 9 Sleep aspects, e.g. sleeps well at night, etc.
19	**NEGATIVE CONSEQUENCES OF CHILD'S ILLNESS (Parental perception of the negative changes in their own child's health and wellbeing post PICU discharge)**	1 Respiratory symptoms 2 Growth and development 3 Diet 4 Sleep 5 Drug therapy 6 Emotion 7 Behaviour 8 Parent's relationship and parental/family wellbeing 9 Health 10 Cognition	1 Respiratory symptoms, e.g. bad cough, chest and lung problems, breathless, etc. 2 Growth and development aspects, e.g. forgot sucking reflex, regression in speech/motor development/toilet training, etc. 3 Dietary aspects, e.g. dietary habit changed, chokes on milk, tube feeding, etc. 4 Sleep aspects, e.g. sleeping pattern disturbed, more sleepy, bad dreams, etc. 5 Drug therapy aspects, e.g. drug withdrawal, amount of drugs, needs nebulisers. 6 Emotional aspects, e.g. crying, wanted cuddles, easily upset, unhappy, etc. 7 Behavioural aspects, e.g. disinterested, not bouncy, attention seeking, etc. 8 Parent's relationship and wellbeing, e.g. affected social life, strain on parents, affected relationship with sibling, difficult to cope, etc. 9 Health aspects, e.g. more prone to illness, generally weak, etc. 10 Cognitive aspects, e.g. behind at school, no understanding of reason for PIC.
20	**NO CONSEQUENCES OF THE CHILD'S ILLNESS (Parental perception of no changes in their own child's health and wellbeing since PICU discharge)**	1 Daily routine 2 Cognition 3 Parent's relationship and/or family wellbeing 4 Growth and development 5 Behaviour 6 Uncertainties 7 Emotion	1 Daily routine aspects, e.g. unchanged, sleeping unchanged, etc. 2 Cognitive aspects, e.g. still bright, still alert, etc. 3 Parent's relationship with child and wellbeing, e.g. parental relationship unchanged, family life unchanged, etc. 4 Growth and development aspects, e.g. normal development, mobility/hearing/speech OK 5 Behavioural aspects, e.g. still the same child, behaviour is the same, etc. 6 Aspects that parents were uncertain if they had changed, e.g. pain threshold, etc. 7 Emotional aspects, e.g. still happy and silly, smiles even when poorly, etc.

Appendix IV (continued) – Definition of themes and categories (Phase I)			
NUD*IST CODE	**THEME (DEFINITION)**	**CATEGORIES**	**DEFINITION OF CATEGORY**
21	**NEUTRAL CONSEQUENCES OF CHILD'S ILLNESS (Changes in a child's health & wellbeing – that cannot be judged as a +ve/-ve change post PICU)**	1 Parent and child relationship 2 Parental values 3 Child's behaviour 4 Health	1 Parent and child relationship, e.g. parent more protective, more watchful, etc. 2 Parental values, e.g. child is most important thing, wanted to spend time with child, values changes following PICU, etc. 3 Child's behaviour, e.g. pain threshold/temperament changed etc. 4 Child's health, e.g. no positive affects on health, no limitations to health, etc.
22	**CONSEQUENCES OF CHILD'S ADDITIONAL ILLNESS (ES) (Parental perception of changes in their own child's health & wellbeing as a result of additional illnesses +ve/ –ve)**	1 Parent and child relationship 2 Daily routine	1 Parent and child relationship, e.g. cosseted child, avoids putting child at risk, etc. 2 Daily routine, e.g. stopped nursery, kept indoors, etc.
23	**REFERENCE TO HEALTH STATE PRE PICU (Parental reference to their own child's health state pre PICU admission)**	1 Respiratory signs and symptoms 2 Sleep 3 Behaviour 4 Emotion 5 Cognition 6 Growth and development 7 Diet 8 Medication 9 Non-respiratory signs and symptoms	1 Respiratory signs and symptoms, e.g. wheezy, breathless, etc. 2 Sleeping pattern, e.g. child's sleeping routine, sleeps all night, etc. 3 Child's behaviour, e.g. determined, placid, content between feeds, etc. 4 Emotional aspects, e.g. cried a lot, no bond with mother, etc. 5 Cognitive aspects, e.g. alert, looking round, not behind at school, etc. 6 Child's growth and development, e.g. skinny, behind in development, walking. 7 Child's diet, e.g. underweight, eats well, tube feeds, total parenteral nutrition. 8 Child's treatment, e.g. drug therapy, etc. 9 Child's non-respiratory signs and symptoms, e.g. vomiting – reflux, etc.
THEMES FOR FUTURE ANALYSIS			
25	**CARE RECEIVED IN HOSPITAL**	1 Treatment on ward 2 Treatment on PICU	
26	**CARE RECEIVED AFTER PICU DISCHARGE**	1 Visits to GP 2 Specialist referral 3 DGH follow up 4 Additional hospital admission 5 Further investigations and monitoring 6 Drug treatment at home 7 Visits from outreach nurse 8 Visit from health visitor	

Appendix V – Thematic content analysis matrix (Phase I)			
THEME (TOTAL NUMBER OF RESPONSES)		**CATEGORIES**	**NUMBER OF RESPONSES (NUMBER OF PARENTS)**
1	**BASE DATA**	1 Parent	NA
		2 Child	NA
2	**MEDICAL HISTORY (286)**	1 Age of child	24 (21)
		2 Length of PICU stay	27 (21)
		3 Previous hospital admissions	17 (13)
		4 Previous PICU admissions	13 (10)
		5 LOS at DGH pre PICU	15 (11)
		6 LOS at DGH post PICU	19 (13)
		7 Total length of hospital stay	4 (4)
		8 Maternity history	27 (12)
		9 Pathway of care to PICU	91 (20)
		10 Pathway of care from PICU	7 (7)
		11 Reason for previous/subsequent admission	15 (9)
		12 Time at home out of hospital	8 (4)
		13 Place of previous hospital admission	19 (8)
3	**STRESSORS - FACTORS CAUSING PARENTS STRESS DURING THEIR CHILD'S ILLNESS (388)**	1 Child's condition	85 (17)
		2 Ward environment	11 (7)
		3 PICU environment	44 (11)
		4 Level of health care delivered	64 (10)
		5 Health care professionals communication	28 (7)
		6 Partner relationship	14 (6)
		7 Family relationships	40 (14)
		8 Parental role	10 (6)
		9 Stressors apart from child's illness crisis (external) - none, travel, work	37 (16)
		10 Managing child at home prior to PICU episode	14 (4)
		11 Managing child at home post PICU episode	41 (13)
4	**PARENTAL WELLBEING (208)**	1 Expression of emotions	188 (19)
		2 Feelings of hope	12 (6)
		3 Physical signs and symptoms (parent)	8 (4)

Appendix V (continued) – Thematic content analysis matrix (Phase I)			
THEME (TOTAL NUMBER OF RESPONSES)		**CATEGORIES**	**NUMBER OF RESPONSES (NUMBER OF PARENTS)**
5	**PARENTAL UNDERSTANDING OF CHILD'S ILLNESS (585)**	1 Social advice (PICU illness)	7 (5)
		2 Family advice (PICU illness)	9 (6)
		3 Health care professional advice (PICU illness)	56 (17)
		4 Advice on additional illness (es)	19 (8)
		5 Respiratory symptoms of PICU illness	104 (19)
		6 Other symptoms of PICU illness (non-respiratory)	60 (13)
		7 Symptoms of additional illnesses	28 (9)
		8 Diagnosis of PICU illness	42 (16)
		9 Diagnosis of additional illness(es)	32 (14)
		10 Cause of PICU illness	59 (16)
		11 Cause of additional illness(es)	37 (12)
		12 Duration of symptoms of PICU illness	37 (16)
		13 Duration of symptoms of additional illness(es)	8 (3)
		14 Cure/control of PICU illness	29 (13)
		15 Cure/control of additional illness(es)	20 (8)
		16 Time for child to return to normal	37 (16)
		17 Time for parent to return to normal	1 (1)
6	**COPING WITH THE CHILD'S ILLNESS (215)**	1 Parents seeking information and support	12 (8)
		2 Family support (visiting)	83 (20)
		3 Information sharing from health care professionals	69 (15)
		4 Coming to terms with the situation	23 (9)
		5 Other	12 (5)
		6 Attitude of health care staff	16 (5)
7	**GLOBAL GOOD QUALITY OF LIFE (112)**	1 Emotion	25 (12)
		2 Behaviour	4 (4)
		3 Cognition	13 (5)
		4 Family	21 (13)
		5 Environment	20 (11)
		6 Inter-personal interaction	13 (7)
		7 Health	7 (6)
		8 Nutrition	9 (5)

Appendix V (continued) – Thematic content analysis matrix (Phase I)			
THEME (TOTAL NUMBER OF RESPONSES)		**CATEGORIES**	**NUMBER OF RESPONSES (NUMBER OF PARENTS)**
8	**SPECIFIC GOOD QUALITY OF LIFE (51)**	1 Inter-personal interaction 2 Behaviour 3 Family 4 Emotion 5 Own child's quality of life (parental perception) 6 Diet	9 (5) 4 (3) 16 (7) 4 (4) 16 (9) 2 (2)
9	**GLOBAL POOR QUALITY OF LIFE (51)**	1 Emotion 2 Environment 3 Family 4 Health 5 Inter-personal interaction 6 Diet	6 (4) 9 (6) 12 (10) 4 (2) 16 (9) 4 (2)
10	**SPECIFIC POOR QUALITY OF LIFE (4)**	1 Inter-personal interaction 2 Behaviour	3 (2) 1 (1)
11	**DISABILITIES AND QUALITY OF LIFE (116)**	1 Factors affecting parental perception of quality of life of a disabled child 2 Parental assessment of quality of life in disabled children	72 (15) 44 (19)
12	**FACTORS AFFECTING PARENTAL QUALITY OF LIFE PERCEPTIONS (114)**	1 Experience with children 2 Parental beliefs and values 3 Parental understanding of the quality of life concept	8 (5) 89 (20) 17 (9)
13	**GLOBAL HEALTH (109)**	1 Emotion 2 Behaviour 3 Cognition 4 Inter-personal interaction 5 Growth and development 6 Diet 7 Environment 8 Family 9 Appearance	22 (11) 18 (10) 6 (5) 10 (7) 24 (13) 16 (10) 2 (1) 2 (1) 9 (2)
14	**SPECIFIC HEALTH (198)**	1Emotion 2 Inter-personal interaction 3 Diet 4 Growth and development 5 Own child's health state pre PICU (parental perception) 6 Own child's health state post PICU (parental perception) 7 Behaviour 8 Appearance	12 (6) 3 (3) 7 (5) 12 (6) 71 (20) 88 (19) 4 (3) 1 (1)

Appendix V (continued) – Thematic content analysis matrix (Phase I)			
THEME (TOTAL NUMBER OF RESPONSES)		**CATEGORIES**	**NUMBER OF RESPONSES (NUMBER OF PARENTS)**
15	**GLOBAL UNHEALTHY (92)**	1 Emotion	13 (8)
		2 Environment	6 (3)
		3 Behaviour	37 (15)
		4 Health	4 (3)
		5 Diet	15 (10)
		6 Inter-personal interaction	5 (4)
		7 Appearance	12 (6)
16	**SPECIFIC UNHEALTHY (52)**	1 Emotion	6 (4)
		2 Behaviour	10 (6)
		3 Symptoms of illness/disease characteristics	30 (11)
		4 Diet	2 (2)
		5 Growth and development	1 (1)
		6 Treatment	3 (2)
17	**FACTORS AFFECTING PARENTAL CHILD HEALTH PERCEPTIONS (216)**	1 Experience of healthy children	51 (17)
		2 Parent's values and beliefs	55 (18)
		3 Parental intuition	28 (10)
		4 Experience of ill children	13 (9)
		5 Parental understanding of the concept of health and ill-health	55 (17)
		6 Parent attitudes to health	14 (8)
18	**POSITIVE CONSEQUENCES OF CHILD'S ILLNESS (175)**	1 Diet	23 (12)
		2 Emotion	25 (11)
		3 Health	12 (8)
		4 Behaviour	38 (12)
		5 Cognition	24 (10)
		6 Parent's relationship and wellbeing	14 (9)
		7 Respiratory	19 (9)
		8 Development	16 (10)
		9 Sleep	4 (2)

Appendix V (continued) – Thematic content analysis matrix (Phase I)			
THEME (TOTAL NUMBER OF RESPONSES)		**CATEGORIES**	**NUMBER OF RESPONSES (NUMBER OF PARENTS)**
19	**NEGATIVE CONSEQUENCES OF CHILD'S ILLNESS (339)**	1 Respiratory symptoms	60 (15)
		2 Growth and development	52 (11)
		3 Diet	34 (10)
		4 Sleep	29 (9)
		5 Drug therapy	8 (6)
		6 Emotion	48 (14)
		7 Behaviour	59 (16)
		8 Parent's relationship and wellbeing	28 (8)
		9 Health	19 (12)
		10 Cognition	2 (1)
20	**NO CONSEQUENCES OF THE CHILD'S ILLNESS (77)**	1 Daily routine	17 (12)
		2 Cognition	6 (3)
		3 Parent's relationship and/or family wellbeing	10 (6)
		4 Growth and development	17 (13)
		5 Behaviour	19 (11)
		6 Uncertainties	3 (3)
		7 Emotion	2 (2)
		8.Same child	3 (1)
21	**NEUTRAL CONSEQUENCES OF CHILD'S ILLNESS (118)**	1 Parent and child relationship	65 (18)
		2 Parental values	12 (8)
		3 Child's behaviour	20 (9)
		4 Health	21 (13)
22	**CONSEQUENCES OF CHILD'S ADDITIONAL ILLNESS(ES) (14)**	1 Parent and child relationship	9 (1)
		2 Daily routine	5 (1)
23	**REFERENCE TO HEALTH STATE PRE PICU (97)**	1 Respiratory signs and symptoms	11 (6)
		2 Sleep	13 (10)
		3 Behaviour	14 (8)
		4 Emotion	16 (9)
		5 Cognition	4 (2)
		6 Growth and development	19 (12)
		7 Diet	17 (11)
		8 Medication	2 (1)
		9 Non-respiratory signs and symptoms	1 (1)

Appendix V (continued) – Thematic content analysis matrix (Phase I)		
		THEMES FOR FUTURE ANALYSIS
25	**CARE RECEIVED IN HOSPITAL**	1 Treatment on ward 2 Treatment on PICU
26	**CARE RECEIVED AFTER PICU DISCHARGE**	1 Visits to GP 2 Specialist referral 3 DGH follow up 4 Additional hospital admission 5 Further investigations and monitoring 6 Drug treatment at home 7 Visits from outreach nurse 8 Visit from HV

Appendix VI – Clinician generated index (draft)

YOUR CHILD'S HEALTH AND WELLBEING

Dear parent/carer

It would be most helpful to the staff on the children's intensive care unit (PICU/Ward 2) if you could spend a few minutes of your time completing this questionnaire about your child's current health and wellbeing.

Information from this questionnaire will be used to improve the delivery and quality of care that children and parents receive on the children's intensive care unit. Additionally, results from this questionnaire will give the staff on the children's intensive care unit a clearer picture of the impact of their care on the child's health and wellbeing following their discharge. This will help the staff to monitor and evaluate the care and services that they provide in both the short and long term.

Any information that you provide on this questionnaire will be treated confidentially and anonymously.

Please circle the most appropriate response and indicate any comments you wish to make in the spaces provided

SECTION A: YOUR CHILD'S SYMPTOMS

Does your child have a day-time cough?	Yes	No
Does your child have a night-time cough?	Yes	No
Does your child have noisy breathing ("a ruttle")?	Yes	No
Does your child have a wheeze? (i.e. make a whistling noise on breathing out)	Yes	No
Does your child have a stridor? (i.e. make a rough noise on breathing in)	Yes	No
Is your child out of breath when still (inactive)?	Yes	No
Is your child breathless when active, e.g walking, running	Yes	No

How fast does your child breathe in a minute /per minute

How would you describe your child's respiratory symptoms?

Mild
Moderate
Severe

Do any noises come from your child's chest during breathing? Yes No

Please describe what these noises sound like

Does your child have any chest deformities, e.g. a pigeon chest, Yes No

If yes, which one(s)?

Pigeon chest
Harrison sulci (def)
Other

How much does your child weigh? /kgs

How tall is your child? /cms
/inches

Is your child's growth normal for his/her age? Yes No

Does your child have any developmental problems? Yes No

If yes, what are they?

Hearing
Speech
Sight
Motor
Understanding
Other.....

SECTION B EFFECT OF YOUR CHILD'S SYMPTOMS ON THEIR DAILY ACTIVITIES

Does your child's respiratory symptoms limit any of the following activities?
(Please indicate N/A if not applicable)

	Yes, limited a lot	Yes, limited a little	No, not limited at all
Eating			
Drinking			
Sitting			
Crawling			
Walking			
Running around at playtime			
Playing sports			
Playing with toys			
Sleeping			
Keeping up with other children			

Does your child have any behavioural problems? (e.g. tantrums) Yes No

If yes, please describe:

Does your child have any emotional problems? (e.g. temprament - clingy, withdrawn) Yes No

If yes, please describe:

Has your child's respiratory symptoms affected attendance at nursery or school? Yes No

If yes, how often?

Every week
Every 2 weeks
Every 3-4 weeks
Every 2-3 months
Every 6 months
Not at all

SECTION C YOUR CHILD'S MEDICATION HISTORY

What medicines is your child taking at the moment?

Name of medicine:	Name of medicine:	Name of medicine:
How much:	How much:	How much:
How often:	How often:	How often:
Which route: (e.g. swallowed, inhaled)	Which route:	Which route:

What medicines have been prescribed by the hospital doctor/GP for your child?

Name of medicine:	Name of medicine:	Name of medicine:
How much:	How much:	How much:
How often:	How often:	How often:
Which route: (e.g. swallowed, inhaled)	Which route:	Which route:

Are there any problems with your child taking their medicine? Yes No

If yes, please describe how:

SECTION D YOUR CHILD'S USE OF HEALTH CARE SERVICES

In the last month, how often have you consulted your GP for any reason related to your child's health? □□

In the last month, how often have you consulted your GP because of your child's respiratory symptoms? □□

In the last month, how often have you consulted your nurse at the GP's surgery for any reason related to your child's health? □□

In the last month, how often have you consulted the nurse at the GP's surgery because of your child's respiratory symptoms? □□

In the last month, how often have you taken your child to the A & E department (casualty) for treatment? □□

In the last month, how often have you taken your child to the A & E department (casualty) for the treatment of respiratory symptoms? □□

How often has your child been admitted to hospital for any treatment in the last month? □□

How often has your child been admitted to hospital for the treatment of respiratory symptoms in the last month? □□

SECTION E YOUR HOME AND FAMILY CIRCUMSTANCES

Who lives with your child?

Name: Relationship: Age:	Name: Relationship: Age:	Name: Relationship: Age:
Name: Relationship: Age:	Name: Relationship: Age:	Name: Relationship: Age:

How many brothers and sisters does your child have? □

What birth order is your child (eldest or youngest)?

First
Second
Third
Fourth
Other .....

What type of accommodation does your child live in?

Flat (__ floor)
Terraced house
Semi-detached
Detached
Bungalow

Does your accommodation have a garden? Yes No

How many rooms are there? ☐

What type of heating is there?

None
Open fires
Storage heaters
Central heating

Does anybody smoke in the family? Yes No

Which family members smoke in the house?

None
Mother
Father
Siblings
Other(s).....

What pets are there in the house?

How often in the past month has your child's illness/emotional state interfered with normal social activities within the family or with friends?

Not at all
Slightly
Moderately
Quite a bit
Extremely

Are you coping with managing your child's illness? Yes No

If no, please describe the reasons why this is difficult

Do you or your family have any concerns or anxieties about your child's illness or treatment? Yes No
If yes, what are they?

How would you describe your child's health at the moment?

Excellent
Very good
Good
Fair
Poor

'Original in colour'

Appendix VII – Retrospective PICQoL questionnaire main study (Phase II)

(Note: The font has been reduced and the margins altered to conform to the thesis margin requirements; the original questionnaire used 1cm margins and size 12 font and was printed on both sides of the page)

ID: □□□

PICQoL (Paediatric Intensive Care Quality of Life) Questionnaire

This questionnaire asks about your child's current health and wellbeing since their discharge from the children's general intensive care unit, also known as PICU. It is important for the doctors and nurses on the PICU to monitor and evaluate the care that they give to children and their families and the services that they provide, both in the short-tem and long-term. The information that you provide in this questionnaire will help doctors and nurses to improve the delivery and quality of care given to children, parents and families on the PICU.

Your answers are important. Any information you provide will not be shared with anyone and your responses will be confidential. Certain questions may look alike, but each one is asking for different information. Please complete each question, your opinions are very important. Please return your questionnaire to Angela Grange in the stamped-addressed envelope provided. Further information is available about this study; please contact Angela Grange (contact details are on Page 11).

SECTION 1: YOUR CHILD'S GENERAL HEALTH

Please time how long it takes you to complete this questionnaire

1.1 At the moment, how would you describe your child's health?

□ Excellent □ Very good □ Good □ Fair □ Poor

1.2 Compare your child's health now with their health when they were well before (the last) PICU admission by choosing <u>one</u> of the responses below:

My child's health is ***much better now*** than before PICU admission □

My child's health is ***somewhat better now*** than before PICU admission □

My child's health is ***the same now*** as before PICU admission □

My child's health is ***somewhat worse now*** than before PICU admission □

My child's health is ***much worse now*** than before PICU admission □

1.3 Has your child been acutely ill (e.g. poorly, broken bones, accident) in the <u>last two weeks</u>?

□ Yes □ No

If **Yes,** please describe below the symptoms of this illness? *E.g. cough, cold, vomiting, diarrhoea, stomach pain, sore throat, etc.*

……
……
……
……
……
……
……

1.4 Here is list of questions asking you to think about your child's everyday activities in the <u>past two weeks</u>. Indicate how true each statement is of your child in comparison to other healthy children the same age as your child, by circling <u>one response only</u> for each statement, where:

0	=	***much less***	than other children the same age
1	=	***somewhat less***	than other children the same age
2	=	***about the same***	as other children the same age
3	=	***somewhat more***	than other children the same age
4	=	***much more***	than other children the same age
NA	=	***not applicable***	my child is not old enough to do this activity yet

Note: *If your child was born prematurely, use your child's corrected gestational age, not their chronological age to compare against a healthy child. For example, if your child was born 3 months premature and is now chronologically aged 9 months, compare your child to a child aged 6 months (corrected gestational age).*

	much less than other children	*somewhat less than other children*	*about the same as other children*	*somewhat more than other children*	*much more than other children*	*NA*
My child moves around *(E.g. sits up/rolls over/crawls/takes steps)*	0	1	2	3	4	NA
My child walks or runs	0	1	2	3	4	NA
My child grasps or picks things up	0	1	2	3	4	NA
My child holds & carries things	0	1	2	3	4	NA
My child's height is	0	1	2	3	4	NA
My child's weight is	0	1	2	3	4	NA
My child's vision is	0	1	2	3	4	NA
My child's hearing is	0	1	2	3	4	NA
My child's speech is *(E.g. coos, babbles, talks)*	0	1	2	3	4	NA
My child feels pain	0	1	2	3	4	NA
My child is anxious	0	1	2	3	4	NA
My child is sad	0	1	2	3	4	NA
My child is happy	0	1	2	3	4	NA
My child is angry	0	1	2	3	4	NA
My child learns	0	1	2	3	4	NA
My child pays attention	0	1	2	3	4	NA
My child understands	0	1	2	3	4	NA
My child plays on his/her own	0	1	2	3	4	NA
My child plays with siblings	0	1	2	3	4	NA
My child plays with friends	0	1	2	3	4	NA
My child cries	0	1	2	3	4	NA
My child laughs	0	1	2	3	4	NA
My child is naughty	0	1	2	3	4	NA
My child behaves him/herself	0	1	2	3	4	NA
My child is tired	0	1	2	3	4	NA
My child is quiet	0	1	2	3	4	NA
My child's tantrums are	0	1	2	3	4	NA

1.5 We are interested in how you saw your child's PICU illness. Indicate how much you feel that the following symptoms were part of the illness that resulted in your child's admission to PICU:

	All of the time	*Frequently*	*Occasionally*	*Never*
Pain or discomfort.....	☐	☐	☐	☐
Feeling sick or vomiting.....	☐	☐	☐	☐
Breathlessness.....	☐	☐	☐	☐
Loss of appetite.....	☐	☐	☐	☐
Lack of energy.....	☐	☐	☐	☐
Sore or sticky eyes.....	☐	☐	☐	☐
Wheeziness	☐	☐	☐	☐
Cough.....	☐	☐	☐	☐
Diarrhoea.....	☐	☐	☐	☐
Sleep difficulties.....	☐	☐	☐	☐
Loss of balance.....	☐	☐	☐	☐
Loss of strength.....	☐	☐	☐	☐
Temperature or fever.....	☐	☐	☐	☐
Runny nose or snuffles.....	☐	☐	☐	☐

1.6 We are interested in your own personal views (not those of your doctor or family) as to how you saw the illness that resulted in your child's admission to PICU. Indicate how much you agree or disagree with the following statements about your child's PICU illness:

	Strongly agree	*Agree*	*Neither agree nor disagree*	*Disagree*	*Strongly Disagree*
A germ or virus caused my child's PICU illness.....	☐	☐	☐	☐	☐
Diet played a major role in causing my child's PICU illness..... ...	☐	☐	☐	☐	☐
Pollution of the environment caused my child's PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness was hereditary – it runs in my family.....	☐	☐	☐	☐	☐
It was just by chance that my child became ill.....	☐	☐	☐	☐	☐
Stress was a major factor in causing my child's PICU illness..... ...	☐	☐	☐	☐	☐
My child's PICU illness was largely due to his/her own behaviour.....	☐	☐	☐	☐	☐
Other people played a large role in causing my child's PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness was caused by poor medical care in the past.....	☐	☐	☐	☐	☐
My child's state of mind played a major part in causing his/her PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness would last a short time..... ...	☐	☐	☐	☐	☐
My child's PICU illness was likely to be permanent rather than temporary..... ...	☐	☐	☐	☐	☐
My child's PICU illness would last for a long time.....	☐	☐	☐	☐	☐

	Strongly agree	*Agree*	*Neither agree nor disagree*	*Disagree*	*Strongly Disagree*
My child's PICU illness was a serious condition.....	☐	☐	☐	☐	☐
My child's PICU illness has had major consequences on his/her life..... ...	☐	☐	☐	☐	☐
My child's PICU illness has become easier to live with.....	☐	☐	☐	☐	☐
My child's PICU illness has not had much effect on his/her life..... ...	☐	☐	☐	☐	☐
My child's PICU illness has strongly affected the way others see him/her.....	☐	☐	☐	☐	☐
My child's PICU illness had serious economic and financial consequences.....	☐	☐	☐	☐	☐
My child's PICU illness has strongly affected the way I see him/her as a person.	☐	☐	☐	☐	☐
My child's PICU illness would improve with time.....	☐	☐	☐	☐	☐
There was a lot which my child could do to control his/her symptoms.....	☐	☐	☐	☐	☐
There was very little that could be done to improve my child's PICU illness.....	☐	☐	☐	☐	☐
Treatment would be effective in curing my child's PICU illness.....	☐	☐	☐	☐	☐
My child's recovery from PICU illness was largely dependent on chance or fate...	☐	☐	☐	☐	☐
What my child did could determine whether his/her PICU illness got better or worse.. ...	☐	☐	☐	☐	☐

1.7 How often during the <u>past two weeks</u> has your child's health:]

	Very often	*Quite often*	*Occasionally*	*Almost never*	*Never*
Interrupted everyday family activities *(E.g. mealtimes)*	☐	☐	☐	☐	☐
Stopped you from going out at short notice	☐	☐	☐	☐	☐
Caused tension or arguments in your home	☐	☐	☐	☐	☐
Caused you to cancel or change plans at the last minute at home or work	☐	☐	☐	☐	☐
Stopped you from going on a family outing	☐	☐	☐	☐	☐
Stopped you/your partner going to work (*Please tick the box if this question is not applicable* ☐)	☐	☐	☐	☐	☐

'Original in colour'

1.8 During the <u>past two weeks</u> how much have you been worried about your child's physical health?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.9 During the <u>past two weeks</u> how much have you been worried about your child's emotional state?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.10 During the <u>past two weeks</u> how much have you been worried about your child's behaviour?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.11 During the <u>past two weeks</u> how much have you been worried about your child's learning abilities?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.12 Describe how you <u>feel right now</u> about <u>your child's health and wellbeing</u> by placing a circle around the most appropriate number to the right of each statement.

	Not at all	*Somewhat*	*Moderately*	*Very much*
I feel calm.....	1	2	3	4
I am tense.....	1	2	3	4
I feel upset.....	1	2	3	4
I am relaxed.....	1	2	3	4
I feel content.....	1	2	3	4
I am worried.....	1	2	3	4

SECTION 2: YOUR CHILD'S RESPIRATORY HEALTH

2.1 How would you describe your child's breathing now?

☐ Excellent ☐ Very good ☐ Good ☐ Fair ☐ Poor

2.2 Compare your child's level of breathing now with their level of breathing when they were well before (the last) PICU admission by choosing <u>one</u> of the responses below:

My child's breathing is ***much better now*** than before PICU admission ☐

My child's breathing is ***somewhat better now*** than before PICU admission ☐

My child's breathing is ***the same now*** as before PICU admission ☐

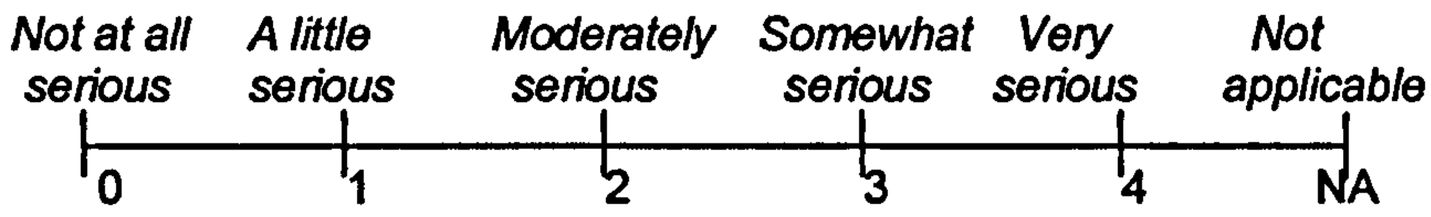
My child's breathing is ***somewhat worse now*** than before PICU admission ☐

My child's breathing is ***much worse now*** than before PICU admission ☐

2.3 a) i) How often has your child experienced a day-time cough in the <u>past two weeks</u>?
(Please tick ONE box only)

Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all ☐

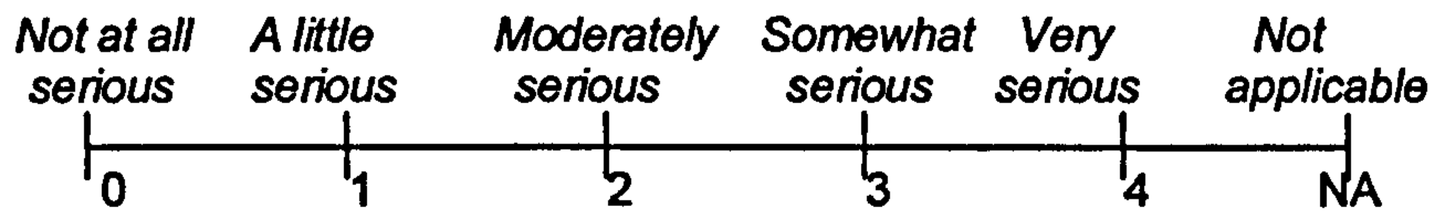
ii) How serious do you think this day-time cough was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



b) i) How often has your child experienced a night-time cough in the <u>past two weeks</u>? *(Please tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

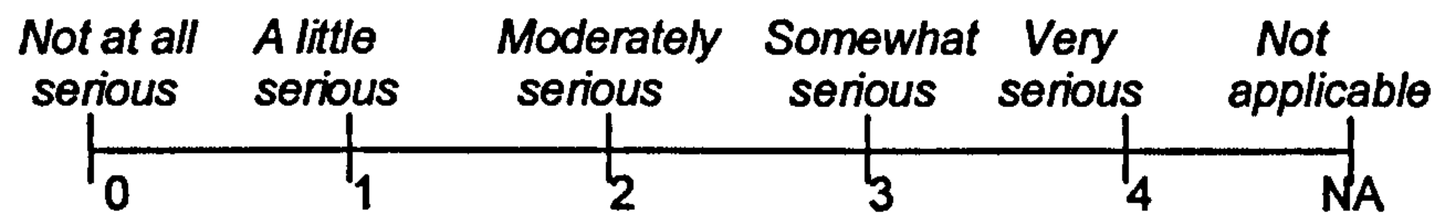
ii) How serious do you think this night-time cough was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



c) i) How often has your child experienced making noisy sounds on breathing in (*a stridor*) in the <u>past two weeks</u>? *(Please tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

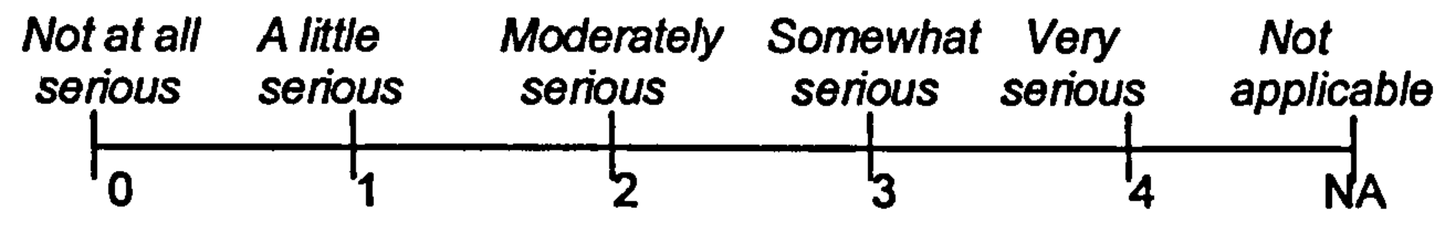
ii) How serious do you think these noisy sounds on breathing in (*a stridor*) were in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



d) i) How often has your child experienced making noisy sounds on breathing out (*a wheeze*) in the <u>past two weeks</u>? *(Please tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

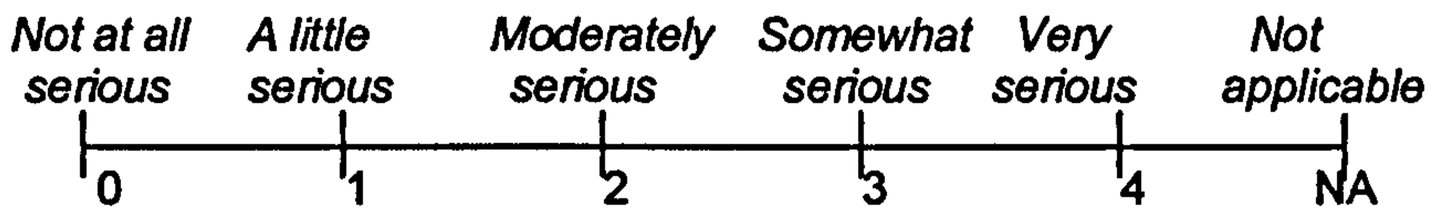
ii) How serious do you think these noisy sounds on breathing out (a wheeze) were in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



e) i) How often has your child experienced breathing at a faster rate than normal in the <u>past two weeks</u>? *(Please tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

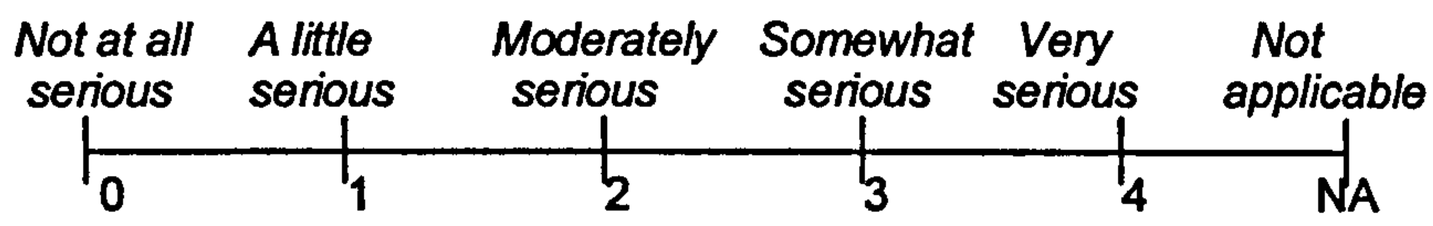
ii) How serious do you think this breathing at a faster rate than normal was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



f) i) How often has your child experienced breathing at a slower rate than normal in the <u>past two weeks</u>? *(Please tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

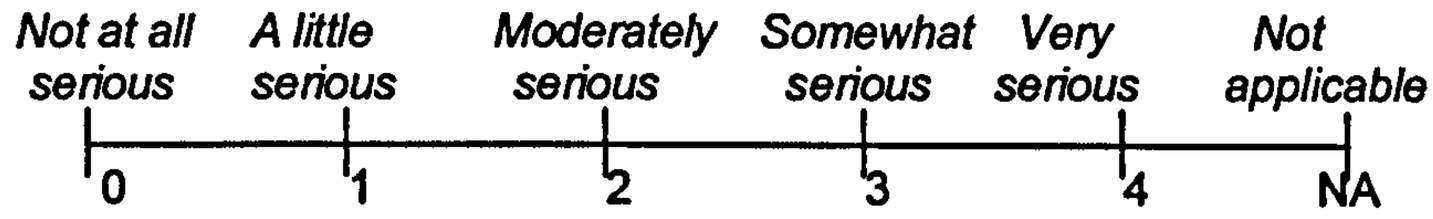
ii) How serious do you think this breathing at a slower rate than normal was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



g) i) How often has your child experienced breathlessness (being out of breath) when still or inactive and resting in the <u>past two weeks</u>? *(tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

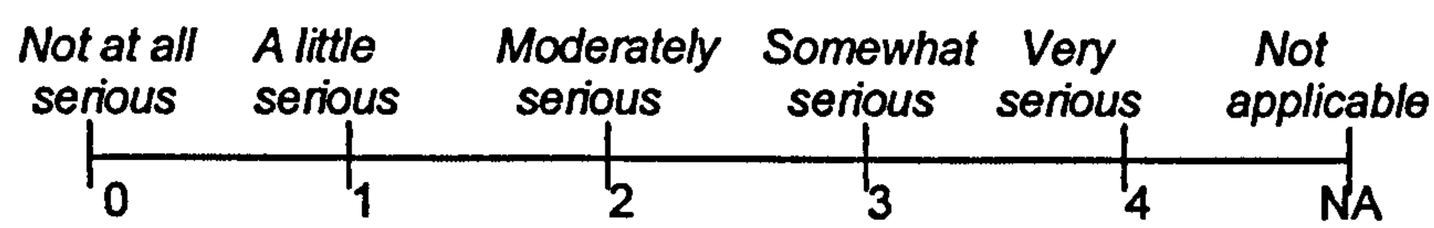
ii) How serious do you think this breathlessness when still or inactive and resting was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



h) i) How often has your child experienced breathlessness (being out of breath) when active, e.g. playing, crawling, walking, running, in the <u>past two weeks</u>? *(tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

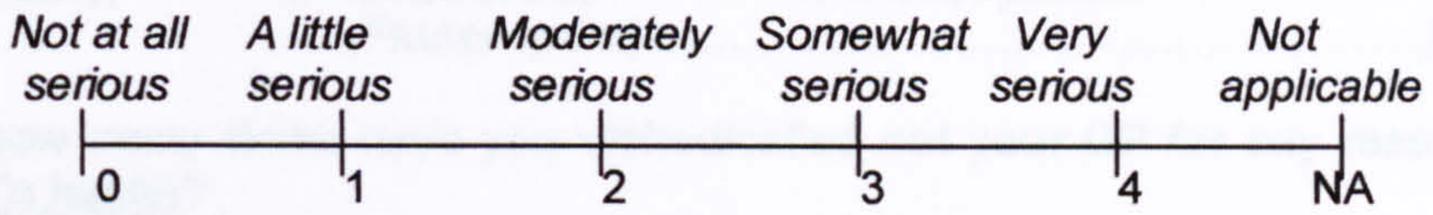
ii) How serious do you think this breathlessness when active was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



i) i) How often has your child had a runny nose or snuffles in the <u>past two weeks</u>? *(tick ONE box only)*

Every day	Every 2-3 days	Every week	Every 2 weeks	Not at all
☐	☐	☐	☐	☐

ii) How serious do you think this runny nose or snuffles were in the past two weeks?
(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)



2.4 Has your child been limited in any of the following everyday activities because of breathing (respiratory) problems in the past two weeks (*Please tick ONE box per activity)?*

		Not limited at all	*Sometimes limited*	*Often limited*	*Always limited*
a)	Eating/feeding	☐	☐	☐	☐
b)	Drinking	☐	☐	☐	☐
c)	Moving around, *(E.g. sitting rolling over/crawling walking)*	☐	☐	☐	☐
d)	Sleeping	☐	☐	☐	☐
e)	Playing	☐	☐	☐	☐
f)	Communicating *(E.g. cooing/ babbling/ talking)*	☐	☐	☐	☐

SECTION 3: FACTS ABOUT YOUR CHILD

3.1 Is your child?

☐ Male ☐ Female

3.2 Was your child born?

☐ First (eldest) ☐ Second ☐ Third ☐ Fourth ☐ Other
(*Please specify*.....)

3.3 Was your child born prematurely?

☐ Yes ☐No

If **Yes,** at what age (in weeks) was your child born *(e.g. 26 weeks)*? ☐☐ weeks

3.4 What is your child's date of birth?

☐☐ / ☐☐ / ☐☐
Day Month Year

3.5 How many times has your child been admitted to hospital as an in-patient before their (last) PICU admission? *(Do not count the time your child was in hospital while being born)*

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.6 If your child was admitted to hospital as an in-patient before their (last) PICU admission, what was the main reason for this?

☐ Breathing (respiratory) problem ☐ Born prematurely ☐ Other illness *(Please specify.....)* ☐ Not applicable

3.7 How many times has your child been admitted to hospital as an in-patient since their (last) PICU admission?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.8 If your child was admitted to hospital as an in-patient since their (last) PICU admission, what was the main reason for this?

☐ Breathing (respiratory) problem ☐ Other illness *(Please specify.....)* ☐ Not applicable

3.9 In the last month, how many times have you visited/called out your GP for any reason related to your child's health?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.10 In the last month, how many times have you visited/called out your GP because of a problem with your child's breathing?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.11 Has a nurse, doctor or other health care professional ever told you that your child has any of the following chest or lung conditions? *(Please tick ONE box per line)*

	Yes	*No*	*Don't know*
Asthma	☐	☐	☐
Chronic respiratory, lung or breathing problem *not asthma*	☐	☐	☐
Cystic fibrosis	☐	☐	☐

3.12 Has a nurse, doctor or other health care professional ever told you that your child has any of the following conditions? *(Please tick ONE box per line)*

	Yes	*No*	*Don't know*
Anxiety problems	☐	☐	☐
Behavioural problems	☐	☐	☐
Cancer	☐	☐	☐
Cerebral palsy	☐	☐	☐
Chest abnormality (e.g. pigeon chest)	☐	☐	☐
Chronic allergies or sinus trouble	☐	☐	☐
Chronic orthopaedic, bone or joint problems	☐	☐	☐
Chronic rheumatic disease	☐	☐	☐
Developmental delay	☐	☐	☐
Diabetes	☐	☐	☐
Digestive (eating and drinking or gut) problems, e.g. reflux	☐	☐	☐
Down's syndrome	☐	☐	☐
Epilepsy (seizure disorder) or fits	☐	☐	☐
Hearing impairment or deafness	☐	☐	☐
Heart problems	☐	☐	☐
Immune problems	☐	☐	☐
Kidney/bladder problems	☐	☐	☐
Learning problems	☐	☐	☐
Liver problems	☐	☐	☐
Neurological (brain/nervous system) problems	☐	☐	☐
Sleep disturbance	☐	☐	☐
Speech or language problems	☐	☐	☐
Vision problems	☐	☐	☐
Any other health problem*	☐	☐	☐

(**Please describe*.....)

3.13 Does your child receive regular treatment and/or medicine?

☐ Yes ☐ No

If **Yes**, please describe below what this regular treatment is and/or the **name**, **amount** and **how often** your child takes any medicines:.....
.....
.....

.....
.....
.....
.....
.....
.....

3.14 Does your child receive oxygen therapy?

□Yes □ No

If **Yes**, a) How often is the oxygen given?

□ Continuously □ Intermittently

b) How much oxygen does your child get in litres/minute during the day and night?

□.□/litres/minute in the day □.□/litres/minute at night

SECTION 4: FACTS ABOUT YOU & YOUR FAMILY

4.1 Are you?

□ Male □ Female

4.2 What is your date of birth?

□□ / □□ / □□

Day Month Year

4.3 Which one of the following best describes your ethnic origin?

□ White
□ Black – Caribbean
□ Black – African
□ Black – other *(Please specify .....)*
□ Indian
□ Pakistani
□ Bangladeshi
□ Chinese
□ Other *(Please specify .....)*

4.4 Which one of the following best describes your marital status?

□ Married □ Living together as a couple □ Widowed □ Divorced or separated □ Single

4.5 Which one of the following best describes your relationship to your child?

□ Biological parent □ Step-parent □ Adoptive parent □ Foster parent □ Other (*Please state.....*)

4.6 Has your work status changed since your child was discharged from PICU?

□ Yes □ No

If **Yes**, please state how it has changed? *(E.g. I now work part-time, I now work full-time, I now do not work because of my child's health problems)*.....
.....
.....
.....

'Original in colour'

4.7 What is the highest grade of educational qualification that you have completed?

☐	☐	☐	☐	☐
No formal education	GCSE or O level	A level	Professional qualification	Degree or or higher

4.8 Do you or anyone in your family smoke in your home?

☐ Yes ☐ No

4.9 Do you have any pets in your home?

☐ Yes ☐ No

If **Yes**, please specify type:.....

4.10 What is today's date?

☐☐ / ☐☐ / ☐☐

Day Month Year

SECTION 5: YOUR VIEWS ABOUT THIS QUESTIONNAIRE

5.1 How many minutes (approximately) did it take you to complete this questionnaire?

☐☐ minutes

5.2 How easy did you find it to complete this questionnaire?

☐	☐	☐	☐	☐
Very easy	Quite easy	Neither easy nor difficult	Quite difficult	Difficult

If you have any comments about this questionnaire or your child's health and care in general, write them below:

COMMENTS:

Please return your questionnaire to Angela Grange
C/o Ward 2
Jubilee Wing
Leeds General Infirmary
Great George Street
Leeds LS1 3EX
Tel: 0113 3923220

Thank you for your participation

'Original in colour'

Appendix VIII – Prospective PICQoL questionnaire main study (Phase II)

(Note: The font has been reduced and the margins altered to conform to the thesis margin requirements; the original questionnaire used 1cm margins and size 12 font and was printed on both sides of the page)

ID: □□□

PICQoL (Paediatric Intensive Care Quality of Life) Questionnaire

This questionnaire asks about your child's current health and wellbeing before and during their admission to the children's general intensive care unit, also known as PICU. It is important for the doctors and nurses on the PICU to monitor and evaluate the care that they give to children and their families and the services that they provide, both in the short-tem and long-term. The information that you provide in this questionnaire will help doctors and nurses to improve the delivery and quality of care given to children, parents and families on the PICU.

Your answers are important. Any information you provide will not be shared with anyone and your responses will be confidential. Certain questions may look alike, but each one is asking for different information. Please complete each question, your opinions are very important. Please return your questionnaire to Angela Grange in the stamped-addressed envelope provided. Further information is available about this study; please contact Angela Grange (contact details are on Page 11).

SECTION 1: YOUR CHILD'S GENERAL HEALTH

Please time how long it takes you to complete this questionnaire.

1.1 At the moment, how would you describe your child's health?

□ Excellent □ Very good □ Good □ Fair □ Poor

1.2 Here is list of questions asking you to think about your child's everyday activities in the <u>two weeks</u> before he/she became unwell with their PICU illness.

Indicate how true each statement is of your child in comparison to other healthy children the same age as your child, by circling <u>one response only</u> (0, 1, 2, 3, 4 or NA) for each statement, where:

0	=	***much less***	than other children the same age
1	=	***somewhat less***	than other children the same age
2	=	***about the same***	as other children the same age
3	=	***somewhat more***	than other children the same age
4	=	***much more***	than other children the same age
NA	=	***not applicable***	(i.e. my child is not old enough to do these activities)

Note: *If your child was born prematurely, use your child's corrected gestational age, not their chronological age to compare against a healthy child. For example, if your child was born 3 months premature and is now chronologically aged 9 months, compare your child to a child aged 6 months (corrected gestational age).*

	much less than other children	*somewhat less than other*	*about the same as other children*	*somewhat more than other children*	*much more than other children children*	*NA*
My child moves around *(E.g. sits up/rolls over/crawls/takes steps)*	0	1	2	3	4	NA
My child walks or runs	0	1	2	3	4	NA
My child grasps or picks things up	0	1	2	3	4	NA
My child holds & carries things	0	1	2	3	4	NA
My child's height is	0	1	2	3	4	NA

	much less than other children	*somewhat less than other*	*about the same as other children*	*somewhat more than other children*	*much more than other children children*	*NA*
My child's weight is	0	1	2	3	4	NA
My child's vision is	0	1	2	3	4	NA
My child's hearing is	0	1	2	3	4	NA
My child's speech is *(E.g. coos, babbles, talks)*	0	1	2	3	4	NA
My child feels pain	0	1	2	3	4	NA
My child is anxious	0	1	2	3	4	NA
My child is sad	0	1	2	3	4	NA
My child is happy	0	1	2	3	4	NA
My child is angry	0	1	2	3	4	NA
My child learns	0	1	2	3	4	NA
My child pays attention	0	1	2	3	4	NA
My child understands	0	1	2	3	4	NA
My child plays on his/her own	0	1	2	3	4	NA
My child plays with siblings	0	1	2	3	4	NA
My child plays with friend	0	1	2	3	4	NA
My child cries	0	1	2	3	4	NA
My child laughs	0	1	2	3	4	NA
My child is naughty	0	1	2	3	4	NA
My child behaves him/herself	0	1	2	3	4	NA
My child is tired	0	1	2	3	4	NA
My child's tantrums are	0	1	2	3	4	NA

1.3 We are interested in how you see your child's PICU illness. Indicate how much you feel that the following symptoms were part of the illness that resulted in your child's admission to PICU:

	All of the time	*Frequently*	*Occasionally*	*Never*
Pain or discomfort.....	☐	☐	☐	☐
Feeling sick or vomiting.....	☐	☐	☐	☐
Breathlessness.....	☐	☐	☐	☐
Loss of appetite.....	☐	☐	☐	☐
Lack of energy.....	☐	☐	☐	☐
Sore or sticky eyes.....	☐	☐	☐	☐
Wheeziness	☐	☐	☐	☐
Cough.....	☐	☐	☐	☐
Diarrhoea.....	☐	☐	☐	☐
Sleep difficulties.....	☐	☐	☐	☐
Loss of balance.....	☐	☐	☐	☐
Loss of strength.....	☐	☐	☐	☐
Temperature or fever.....	☐	☐	☐	☐
Runny nose or snuffles.....	☐	☐	☐	☐

1.4 We are interested in your own personal views (not those of your doctor or family) as to how you see the illness that resulted in your child's admission to PICU. Indicate how much you agree or disagree with the following statements about your child's PICU illness:

	Strongly agree	*Agree*	*Neither agree nor disagree*	*Disagree*	*Strongly Disagree*
A germ or virus caused my child's PICU illness.....	☐	☐	☐	☐	☐
Diet played a major role in causing my child's PICU illness..... ...	☐	☐	☐	☐	☐
Pollution of the environment caused my child's PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness is hereditary – it runs in my family.....	☐	☐	☐	☐	☐
It was just by chance that my child became ill.....	☐	☐	☐	☐	☐
Stress was a major factor in causing my child's PICU illness..... ...	☐	☐	☐	☐	☐
My child's PICU illness is largely due to his/her own behaviour.....	☐	☐	☐	☐	☐
Other people played a large role in causing my child's PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness was caused by poor medical care in the past.....	☐	☐	☐	☐	☐
My child's state of mind played a major part in causing his/her PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness will last a short time	☐	☐	☐	☐	☐
My child's PICU illness is likely to be permanent rather than temporary..... ...	☐	☐	☐	☐	☐
My child's PICU illness will last for a long time.....	☐	☐	☐	☐	☐
My child's PICU illness is a serious condition.....	☐	☐	☐	☐	☐
My child's PICU illness has had major consequences on his/her life..... ...	☐	☐	☐	☐	☐
My child's PICU illness has become easier to live with.....	☐	☐	☐	☐	☐
My child's PICU illness has not had much effect on his/her life..... ...	☐	☐	☐	☐	☐
My child's PICU illness has strongly affected the way others see him/her.....	☐	☐	☐	☐	☐
My child's PICU illness has serious economic and financial consequences.....	☐	☐	☐	☐	☐
My child's PICU illness has strongly affected the way I see him/her as a person...	☐	☐	☐	☐	☐
My child's PICU illness will improve with time.....	☐	☐	☐	☐	☐

	Strongly agree	*Agree*	*Neither agree nor disagree*	*Disagree*	*Strongly Disagree*
There is a lot which my child can do to control his/her symptoms.....	☐	☐	☐	☐	☐
There is very little that can be done to improve my child's PICU illness.....	☐	☐	☐	☐	☐
Treatment will be effective in curing my child's PICU illness.....	☐	☐	☐	☐	☐
My child's recovery from PICU illness is largely dependent on chance or fate.....	☐	☐	☐	☐	☐
What my child does can determine whether his/her PICU illness gets better or worse.....	☐	☐	☐	☐	☐

1.5 During the <u>two weeks</u> before your child became unwell with their PICU illness, how often has your child's health:

	Very often	*Quite often*	*Occasionally*	*Almost never*	*Never*
Interrupted everyday family activities *(E.g. mealtimes)*	☐	☐	☐	☐	☐
Stopped you from going out at short notice	☐	☐	☐	☐	☐
Caused tension or arguments in your home	☐	☐	☐	☐	☐
Caused you to cancel or change plans at home or work	☐	☐	☐	☐	☐
Stopped you from going on a family outing	☐	☐	☐	☐	☐
Stopped you/your partner going to work (*Please tick the box if not applicable* ☐)	☐	☐	☐	☐	☐

1.6 During the <u>two weeks</u> before your child became unwell with their PICU illness, how much have you been worried about their physical health?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.7 During the <u>two weeks</u> before your child became unwell with their PICU illness, how much have you been worried about your child's emotional state?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.8 During the <u>two weeks</u> before you child became unwell with their PICU illness, how much have you been worried about your child's behaviour?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.9 During the <u>two weeks</u> before your child became unwell with their PICU illness, how much have you been worried about your child's learning abilities?

☐ Not at all ☐ A little bit ☐ Some ☐ Quite a bit ☐ A lot

1.10 Describe how you <u>feel right now</u> about <u>your child's health and wellbeing</u> by placing a circle around the most appropriate number to the right of each statement.

	Not at all	Somewhat	Moderately	Very much
I feel calm.....	1	2	3	4
I am tense.....	1	2	3	4
I feel upset.....	1	2	3	4
I am relaxed.....	1	2	3	4
I feel content.....	1	2	3	4
I am worried.....	1	2	3	4

SECTION 2: YOUR CHILD'S RESPIRATORY HEALTH

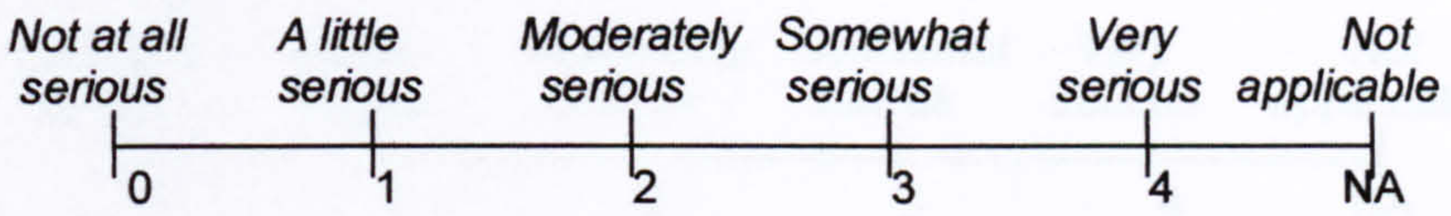
2.1 How would you describe your child's breathing now?

☐ Excellent ☐ Very good ☐ Good ☐ Fair ☐ Poor

2.2 a) i) How often has your child experienced a day-time cough in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

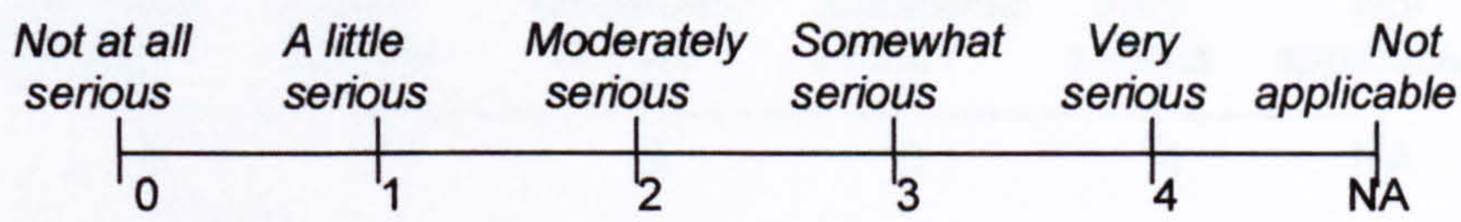
ii) How serious do you think this day-time cough was in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



b) i) How often has your child experienced a night-time cough in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

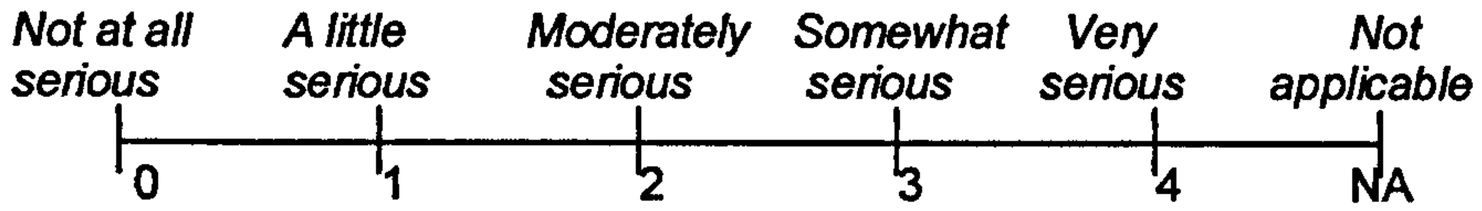
ii) How serious do you think this night-time cough was in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



c) i) How often has your child experienced making noisy sounds on breathing in (*a stridor*) in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

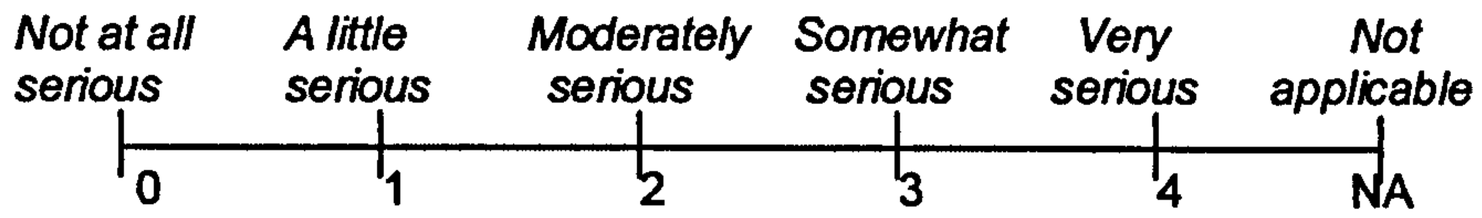
ii) How serious do you think these noisy sounds on breathing in (*a stridor*) were in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



d) i) How often has your child experienced making noisy sounds on breathing out (*a wheeze*) in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

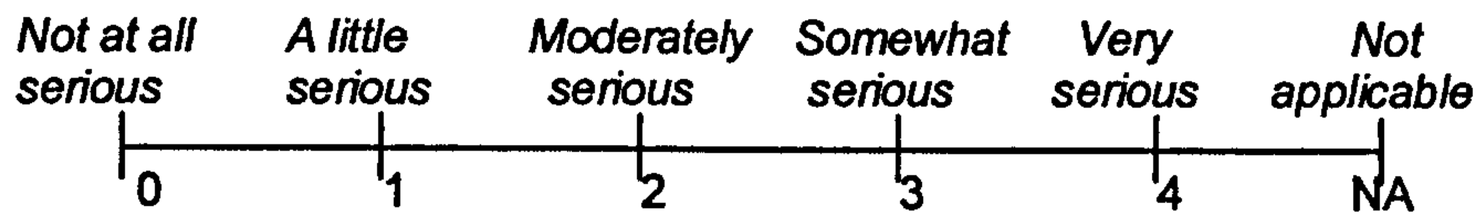
ii) How serious do you think these noisy sounds on breathing out (a wheeze) were in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



e) i) How often has your child experienced breathing at a faster rate than normal in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

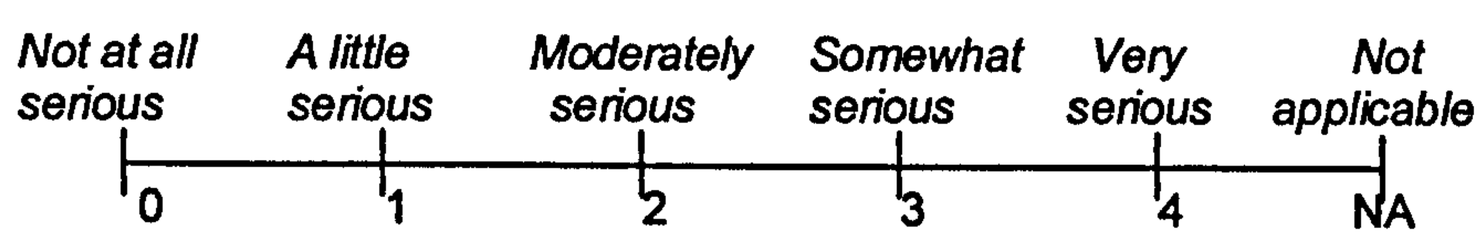
ii) How serious do you think this breathing at a faster rate than normal was in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



f) i) How often has your child experienced breathing at a slower rate than normal in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

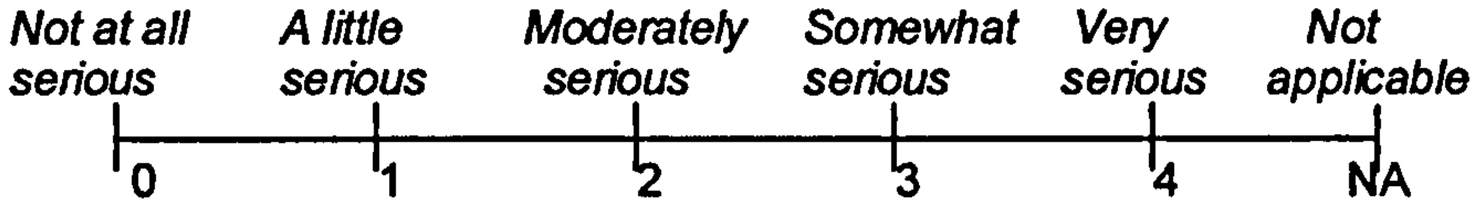
ii) How serious do you think this breathing at a slower rate than normal was in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



g) i) How often has your child experienced breathlessness(being out of breath) when still or inactive and resting in the <u>two weeks</u> before they were unwell with their PICU illness? *(tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

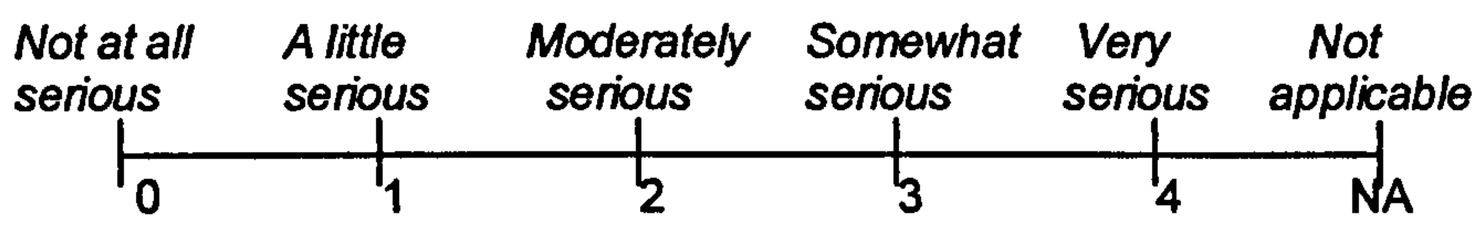
ii) How serious do you think this breathlessness when still or inactive and resting was in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



h) i) How often has your child experienced breathlessness (being out of breath) when active, e.g. playing, crawling, walking, running, in the <u>two weeks</u> before they were unwell with their PICU illness? *(tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

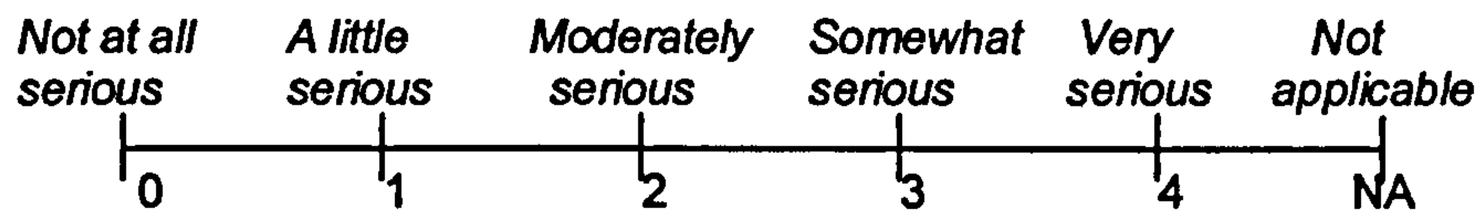
ii) How serious do you think this breathlessness when active was in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



i) i) How often has your child had a runny nose or snuffles in the <u>two weeks</u> before they were unwell with their PICU illness? *(tick ONE box only)*

☐ Every day ☐ Every 2-3 days ☐ Every week ☐ Every 2 weeks ☐ Not at all

ii) How serious do you think this runny nose or snuffles were in the <u>two weeks</u> before they were unwell with their PICU illness? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle NA = not applicable)*



2.3 Has your child been limited in any of the following everyday activities because of breathing (respiratory) problems in the <u>two weeks</u> before they were unwell with their PICU illness? (*Please tick ONE box per activity)?*

		Not limited at all	*Sometimes limited*	*Often limited*	*Always limited*
a)	Eating/feeding	☐	☐	☐	☐
b)	Drinking	☐	☐	☐	☐
c)	Moving around, *(E.g. sitting/rolling over/crawling/walking)*	☐	☐	☐	☐
d)	Sleeping	☐	☐	☐	☐
e)	Playing	☐	☐	☐	☐
f)	Communicating *(E.g. cooing/ babbling/ talking)*	☐	☐	☐	☐

'Original in colour'

SECTION 3: FACTS ABOUT YOUR CHILD

3.1 Is your child?

☐ Male ☐ Female

3.2 a) Was your child born?

☐ First (eldest) ☐ Second ☐ Third ☐ Fourth ☐ Other
(*Please specify*.....)

b) Is your child a

☐ Singleton ☐ Twin ☐ Triplet ☐ Other multiple
(*Please specify*.....)

3.3 Was your child born prematurely?

☐ Yes ☐ No
If **Yes,** at what age (in weeks) was your child born *(e.g. 26 weeks)*? ☐☐ weeks

3.4 What is your child's date of birth?

☐☐ / ☐☐ / ☐☐
Day Month Year

3.5 How many times has your child been admitted to hospital as an in-patient before this PICU admission? *(Do not count the time your child was in hospital while being born)*

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than 5

3.6 If your child was admitted to hospital as an in-patient before this PICU admission, what was the main reason for this?

☐ Breathing (respiratory) problem ☐ Born prematurely ☐ Other illness *(Please specify.....)* ☐ Not applicable

3.7 Is this your child's first admission to PICU?

☐ Yes ☐ No ☐ Don't know

3.8 In the <u>last month</u> (before PICU admission), how many times have you visited/called out your GP for any reason related to your child's health?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than 5

3.9 In the <u>last month</u> (before PICU admission), how many times have you visited/called out your GP because of a problem with your child's breathing?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than 5

3.10 Has a nurse, doctor or other health care professional ever told you that your child has any of the following chest or lung conditions? *(Please tick ONE box per line)*

	Yes	*No*	*Don't know*
Asthma.....	☐	☐	☐
Chronic respiratory, lung or breathing problem *not asthma* .	☐	☐	☐
Cystic fibrosis.....	☐	☐	☐

3.11 Has a nurse, doctor or other health care professional ever told you that your child has any of the following conditions? *(Please tick ONE box per line)*

	Yes	*No*	*Don't know*
Anxiety problems	☐	☐	☐
Behavioural problems	☐	☐	☐
Cancer	☐	☐	☐
Cerebral palsy	☐	☐	☐
Chest abnormality (e.g. pigeon chest)	☐	☐	☐
Chronic allergies or sinus trouble	☐	☐	☐
Chronic orthopaedic, bone or joint problems	☐	☐	☐
Chronic rheumatic disease	☐	☐	☐
Developmental delay	☐	☐	☐
Diabetes	☐	☐	☐
Digestive (eating and drinking or gut) problems, e.g. reflux	☐	☐	☐
Down's syndrome	☐	☐	☐
Epilepsy (seizure disorder) or fits	☐	☐	☐
Hearing impairment or deafness	☐	☐	☐
Heart problems	☐	☐	☐
Immune problems	☐	☐	☐
Kidney/bladder problems	☐	☐	☐
Learning problems	☐	☐	☐
Liver problems	☐	☐	☐
Neurological (brain/nervous system) problems	☐	☐	☐
Sleep disturbance	☐	☐	☐
Speech or language problems	☐	☐	☐
Vision problems	☐	☐	☐
Any other health problem*	☐	☐	☐

(**Please describe*.....)

3.12 Does your child receive regular treatment and/or medicine at home?

☐ Yes ☐ No

If **Yes**, please describe below what this regular treatment is and/or the **name**, **amount** and **how often** your child takes any medicines:.....

.....

.....

.....

.....

.....

.....

.....

.....

.....

3.13 Does your child receive oxygen therapy at home?

☐ Yes ☐ No

If **Yes**, a) How often is the oxygen given?

☐ Continuously ☐ Intermittently

b) How much oxygen does your child get in litres/minute during the day and night?

☐.☐/litres/minute in the day ☐.☐/litres/minute at night

'Original in colour'

SECTION 4: FACTS ABOUT YOU & YOUR FAMILY

4.1 Are you?

□ Male □ Female

4.2 What is your date of birth?

□□ / □□ / □□
Day Month Year

4.3 Which one of the following best describes your ethnic origin?

□ White
□ Black – Caribbean
□ Black – African
□ Black – other *(Please specify .....)*
□ Indian
□ Pakistani
□ Bangladeshi
□ Chinese
□ Other *(Please specify .....)*

4.4 Which one of the following best describes your marital status?

□ Married
□ Living together as a couple
□ Widowed
□ Divorced or separated
□ Single

4.5 Which one of the following best describes your relationship to your child?

□ Biological parent
□ Step-parent
□ Adoptive parent
□ Foster parent
□ Other (*Please state.....*)

4.6i) Which of the following best describes your current work status? *(Please tick ONE box)*

□ Working full-time for 30 hours or more per week
□ Working part-time for less than 30 hours per week
□ Unemployed due to illness or disability
□ Unemployed not due to illness or disability
□ Retired due to illness or disability
□ Retired not due to illness or disability
□ Housewife/home maker
□ Full-time student

4.7ii) Has your work status changed since your child was admitted to PICU?

□ Yes □ No

If **Yes**, please state how it has changed? *(E.g. I now work part-time, I now work full-time, I now do not work because of my child's health problems)*.....
.....
.....
.....

4.8 What is the highest grade of educational qualification that you have completed?

□ No formal education
□ GCSE or O level
□ A level
□ Professional qualification
□ Degree or or higher

4.9 Do you or anyone in your family smoke in your home?

☐ Yes ☐ No

4.10 Do you have any pets in your home?

☐ Yes ☐ No

If **Yes**, please specify type:.....

4.11 What is today's date?

☐☐ / ☐☐ / ☐☐

Day Month Year

SECTION 5: YOUR VIEWS ABOUT THIS QUESTIONNAIRE

5.1 How many minutes (approximately) did it take you to complete this questionnaire?

☐☐ minutes

5.2 How easy did you find it to complete this questionnaire?

☐	☐	☐	☐	☐
Very easy	Quite easy	Neither easy nor difficult	Quite difficult	Difficult

If you have any comments about this questionnaire or your child's health and care in general, write them below:

COMMENTS:

Please return your questionnaire to Angela Grange
C/o Ward 2
Jubilee Wing
Leeds General Infirmary
Great George Street
Leeds LS1 3EX
Tel: 0113 392 3220
Thank you for your participation

Appendix IX - Factor analyses of daily activities items (Phase II)

PAF (1-factor solution) of daily activities items

Communalities

	Initial	Extraction
Level of movement - recoded	.796	.415
Walks or runs - recoded	.809	.383
Grasps or picks things up - recoded	.855	.565
Holds & carries things - recoded	.877	.680
Child's height - recoded	.585	.195
Child's weight - recoded	.669	.156
Child's vision - recoded	.577	.249
Child's hearing - recoded	.686	6.559E-02
Child's speech - recoded	.704	.544
Child feeling pain - recoded	.420	6.436E-02
Child is anxious - recoded	.492	5.206E-02
Child is sad - recoded	.638	.164
Child is happy - recoded	.496	.169
Child is angry - recoded	.702	3.105E-02
Child learns - recoded	.673	.511
Child pays attention - recoded	.702	.124
Child understands - recoded	.681	.478
Child plays on own - recoded	.539	6.490E-02
Child plays with siblings - recoded	.506	.309
Child plays with friends - recoded	.613	.388
Child cries - recoded	.688	9.469E-02
Child laughs - recoded	.563	.299
Child is naughty - recoded	.671	.107
Child behaves - recoded	.500	3.919E-02
Child is tired - recoded	.608	9.231E-02
Child is quiet - recoded	.459	3.806E-02
Child has tantrums - recoded	.576	1.253E-04

Extraction Method: Principal Axis Factoring.

Note: $-02 = 10^{-2}$ so $6.559E\text{-}02 = 0.06559$
$-04 = 10^{-4}$ so $1.253E\text{-}04 = 0.0001253$

Total Variance Explained

Factor	Initial Eigenvalues			Extraction Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	6.890	25.520	25.520	6.277	23.247	23.247
2	3.155	11.686	37.206			
3	2.262	8.379	45.585			
4	1.837	6.802	52.387			
5	1.540	5.704	58.092			
6	1.425	5.279	63.371			
7	1.282	4.747	68.118			
8	1.093	4.050	72.168			
9	.859	3.182	75.350			
10	.826	3.061	78.411			
11	.688	2.549	80.960			
12	.623	2.306	83.267			
13	.563	2.086	85.353			
14	.535	1.982	87.335			
15	.509	1.885	89.220			
16	.450	1.667	90.887			
17	.400	1.480	92.367			
18	.373	1.383	93.750			
19	.319	1.181	94.931			
20	.281	1.041	95.972			
21	.270	1.001	96.973			
22	.210	.777	97.750			
23	.198	.734	98.484			
24	.126	.465	98.950			
25	.117	.433	99.382			
26	.102	.379	99.761			
27	6.453E-02	.239	100.000			

Extraction Method: Principal Axis Factoring.

Factor Matrix[a]

	Factor
	1
Holds & carries things - recoded	.825
Grasps or picks things up - recoded	.751
Child's speech - recoded	.738
Child learns - recoded	.715
Child understands - recoded	.692
Level of movement - recoded	.644
Child plays with friends - recoded	.623
Walks or runs - recoded	.619
Child plays with siblings - recoded	.556
Child laughs - recoded	.547
Child's vision - recoded	.499
Child's height - recoded	.441
Child is happy - recoded	.411
Child is sad - recoded	-.405
Child's weight - recoded	.395
Child pays attention - recoded	.353
Child is naughty - recoded	.327
Child cries - recoded	-.308
Child is tired - recoded	-.304
Child's hearing - recoded	
Child plays on own - recoded	
Child feeling pain - recoded	
Child is anxious - recoded	
Child behaves - recoded	
Child is quiet - recoded	
Child is angry - recoded	
Child has tantrums - recoded	

Extraction Method: Principal Axis Factoring.

a. 1 factors extracted. 4 iterations required.

Note: Factor loading scores less than 0.3 are shown as blank in this table

PAF (2-factor solution) oblique rotation (direct obliminal) of daily activities items

Communalities

	Initial
Level of movement - recoded	.796
Walks or runs - recoded	.809
Grasps or picks things up - recoded	.855
Holds & carries things - recoded	.877
Child's height - recoded	.585
Child's weight - recoded	.669
Child's vision - recoded	.577
Child's hearing - recoded	.686
Child's speech - recoded	.704
Child feeling pain - recoded	.420
Child is anxious - recoded	.492
Child is sad - recoded	.638
Child is happy - recoded	.496
Child is angry - recoded	.702
Child learns - recoded	.673
Child pays attention - recoded	.702
Child understands - recoded	.681
Child plays on own - recoded	.539
Child plays with siblings - recoded	.506
Child plays with friends - recoded	.613
Child cries - recoded	.688
Child laughs - recoded	.563
Child is naughty - recoded	.671
Child behaves - recoded	.500
Child is tired - recoded	.608
Child is quiet - recoded	.459
Child has tantrums - recoded	.576

Extraction Method: Principal Axis Factoring.

Total Variance Explained

Factor	Initial Eigenvalues			Rotation
	Total	% of Variance	Cumulative %	Total
1	6.890	25.520	25.520	6.037
2	3.155	11.686	37.206	3.578
3	2.262	8.379	45.585	
4	1.837	6.802	52.387	
5	1.540	5.704	58.092	
6	1.425	5.279	63.371	
7	1.282	4.747	68.118	
8	1.093	4.050	72.168	
9	.859	3.182	75.350	
10	.826	3.061	78.411	
11	.688	2.549	80.960	
12	.623	2.306	83.267	
13	.563	2.086	85.353	
14	.535	1.982	87.335	
15	.509	1.885	89.220	
16	.450	1.667	90.887	
17	.400	1.480	92.367	
18	.373	1.383	93.750	
19	.319	1.181	94.931	
20	.281	1.041	95.972	
21	.270	1.001	96.973	
22	.210	.777	97.750	
23	.198	.734	98.484	
24	.126	.465	98.950	
25	.117	.433	99.382	
26	.102	.379	99.761	
27	6.453E-02	.239	100.000	

Extraction Method: Principal Axis Factoring.

a. When factors are correlated, sums of squared loadings cannot be added to obtain a total variance.

Structure Matrix

	Factor	
	1	2
Holds & carries things - recoded	.891	
Grasps or picks things up - recoded	.795	
Child learns - recoded	.685	-.358
Child's speech - recoded	.674	-.476
Level of movement - recoded	.663	
Child plays with friends - recoded	.655	
Child understands - recoded	.654	-.376
Walks or runs - recoded	.638	
Child plays with siblings - recoded	.588	
Child's vision - recoded	.548	
Child is naughty - recoded	.489	.311
Child laughs - recoded	.471	-.451
Child's height - recoded	.376	-.373
Child's weight - recoded	.358	
Child pays attention - recoded	.321	
Child's hearing - recoded		
Child cries - recoded		.715
Child is angry - recoded		.710
Child is sad - recoded		.546
Child has tantrums - recoded		.478
Child is happy - recoded	.314	-.477
Child feeling pain - recoded		.462
Child is anxious - recoded		.375
Child is tired - recoded		
Child is quiet - recoded		
Child plays on own - recoded		
Child behaves - recoded		

Extraction Method: Principal Axis Factoring.
Rotation Method: Oblimin with Kaiser Normalization.

Note: Factor loading scores less than 0.3 are shown as blank in this table

Factor Correlation Matrix

Factor	1	2
1	1.000	-.257
2	-.257	1.000

Extraction Method: Principal Axis Factoring.
Rotation Method: Oblimin with Kaiser Normalization.

PAF (3-factor solution) oblique rotation (direct obliminal) of daily activities items

Communalities

	Initial
Level of movement - recoded	.796
Walks or runs - recoded	.809
Grasps or picks things up - recoded	.855
Holds & carries things - recoded	.877
Child's height - recoded	.585
Child's weight - recoded	.669
Child's vision - recoded	.577
Child's hearing - recoded	.686
Child's speech - recoded	.704
Child feeling pain - recoded	.420
Child is anxious - recoded	.492
Child is sad - recoded	.638
Child is happy - recoded	.496
Child is angry - recoded	.702
Child learns - recoded	.673
Child pays attention - recoded	.702
Child understands - recoded	.681
Child plays on own - recoded	.539
Child plays with siblings - recoded	.506
Child plays with friends - recoded	.613
Child cries - recoded	.688
Child laughs - recoded	.563
Child is naughty - recoded	.671
Child behaves - recoded	.500
Child is tired - recoded	.608
Child is quiet - recoded	.459
Child has tantrums - recoded	.576

Extraction Method: Principal Axis Factoring.

Total Variance Explained

Factor	Initial Eigenvalues			Rotation
	Total	% of Variance	Cumulative %	Total
1	6.890	25.520	25.520	6.075
2	3.155	11.686	37.206	3.323
3	2.262	8.379	45.585	1.987
4	1.837	6.802	52.387	
5	1.540	5.704	58.092	
6	1.425	5.279	63.371	
7	1.282	4.747	68.118	
8	1.093	4.050	72.168	
9	.859	3.182	75.350	
10	.826	3.061	78.411	
11	.688	2.549	80.960	
12	.623	2.306	83.267	
13	.563	2.086	85.353	
14	.535	1.982	87.335	
15	.509	1.885	89.220	
16	.450	1.667	90.887	
17	.400	1.480	92.367	
18	.373	1.383	93.750	
19	.319	1.181	94.931	
20	.281	1.041	95.972	
21	.270	1.001	96.973	
22	.210	.777	97.750	
23	.198	.734	98.484	
24	.126	.465	98.950	
25	.117	.433	99.382	
26	.102	.379	99.761	
27	6.453E-02	.239	100.000	

Extraction Method: Principal Axis Factoring.

a. When factors are correlated, sums of squared loadings cannot be added to obtain a total variance.

Structure Matrix

	Factor		
	1	2	3
Holds & carries things - recoded	.896		
Grasps or picks things up - recoded	.807		
Level of movement - recoded	.692		
Child's speech - recoded	.691	-.441	
Child learns - recoded	.672		.374
Walks or runs - recoded	.665		
Child plays with friends - recoded	.649		
Child understands - recoded	.640		.390
Child plays with siblings - recoded	.578		
Child's vision - recoded	.539		
Child is naughty - recoded	.478	.348	
Child laughs - recoded	.467	-.403	
Child's height - recoded	.374	-.336	
Child's weight - recoded	.362		
Child's hearing - recoded			
Child is angry - recoded		.740	
Child cries - recoded		.704	
Child has tantrums - recoded		.518	
Child is sad - recoded		.516	
Child feeling pain - recoded		.443	
Child is happy - recoded		-.430	.405
Child is anxious - recoded		.417	
Child is tired - recoded		.324	
Child behaves - recoded			.560
Child pays attention - recoded			.552
Child is quiet - recoded		.335	.498
Child plays on own - recoded			.441

Extraction Method: Principal Axis Factoring.
Rotation Method: Oblimin with Kaiser Normalization.

Note: Factor loading scores less than 0.3 are shown as blank in this table

PAF 3-factor solution (varimax rotation) of daily activities items

Communalities

	Initial
Level of movement - recoded	.796
Walks or runs - recoded	.809
Grasps or picks things up - recoded	.855
Holds & carries things - recoded	.877
Child's height - recoded	.585
Child's weight - recoded	.669
Child's vision - recoded	.577
Child's hearing - recoded	.686
Child's speech - recoded	.704
Child feeling pain - recoded	.420
Child is anxious - recoded	.492
Child is sad - recoded	.638
Child is happy - recoded	.496
Child is angry - recoded	.702
Child learns - recoded	.673
Child pays attention - recoded	.702
Child understands - recoded	.681
Child plays on own - recoded	.539
Child plays with siblings - recoded	.506
Child plays with friends - recoded	.613
Child cries - recoded	.688
Child laughs - recoded	.563
Child is naughty - recoded	.671
Child behaves - recoded	.500
Child is tired - recoded	.608
Child is quiet - recoded	.459
Child has tantrums - recoded	.576

Extraction Method: Principal Axis Factoring.

Total Variance Explained

Factor	Initial Eigenvalues			Rotation Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	6.890	25.520	25.520	5.576	20.653	20.653
2	3.155	11.686	37.206	2.969	10.996	31.649
3	2.262	8.379	45.585	2.040	7.556	39.205
4	1.837	6.802	52.387			
5	1.540	5.704	58.092			
6	1.425	5.279	63.371			
7	1.282	4.747	68.118			
8	1.093	4.050	72.168			
9	.859	3.182	75.350			
10	.826	3.061	78.411			
11	.688	2.549	80.960			
12	.623	2.306	83.267			
13	.563	2.086	85.353			
14	.535	1.982	87.335			
15	.509	1.885	89.220			
16	.450	1.667	90.887			
17	.400	1.480	92.367			
18	.373	1.383	93.750			
19	.319	1.181	94.931			
20	.281	1.041	95.972			
21	.270	1.001	96.973			
22	.210	.777	97.750			
23	.198	.734	98.484			
24	.126	.465	98.950			
25	.117	.433	99.382			
26	.102	.379	99.761			
27	6.453E-02	.239	100.000			

Extraction Method: Principal Axis Factoring.

Rotated Factor Matrix[a]

	Factor		
	1	2	3
Holds & carries things - recoded	.898		
Grasps or picks things up - recoded	.809		
Level of movement - recoded	.703		
Walks or runs - recoded	.676		
Child's speech - recoded	.644	-.384	
Child plays with friends - recoded	.633		
Child learns - recoded	.605		.379
Child understands - recoded	.568		.397
Child plays with siblings - recoded	.562		
Child is naughty - recoded	.536	.389	
Child's vision - recoded	.534		
Child laughs - recoded	.393	-.337	
Child's weight - recoded	.330		
Child's height - recoded	.316		
Child's hearing - recoded			
Child is angry - recoded		.751	
Child cries - recoded		.678	
Child has tantrums - recoded		.548	
Child is sad - recoded		.470	
Child is anxious - recoded		.440	
Child feeling pain - recoded		.413	
Child is tired - recoded		.337	
Child behaves - recoded			.564
Child pays attention - recoded			.555
Child is quiet - recoded		.393	.465
Child plays on own - recoded			.452
Child is happy - recoded		-.354	.441

Extraction Method: Principal Axis Factoring.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 7 iterations.

Note: Factor loading scores less than 0.3 are shown as blank in this table

'Original in colour'

Appendix X – PICQoL questionnaire (Phase III)

(Note: The font has been reduced and the layout and margins altered to conform to thesis margin requirements; the original questionnaire used 0.5 cm margins and size 12 font and was printed on both sides of the page)

ID: □□□

PICQoL (Paediatric Intensive Care Quality of Life) Questionnaire

This questionnaire asks about your child's current health and wellbeing since their discharge from the children's general intensive care unit, also known as PICU. We appreciate that your child may have been admitted to PICU more than once, we would like you to answer this questionnaire in relation to your child's most recent admission to PICU as a result of a respiratory (breathing) problem. The information that you provide in this questionnaire will help to improve the quality of care given to patients and their families on the PICU.

Please complete each question, your opinions are very important. Certain questions may look alike, but each one is asking for different information. Any information you provide will not be shared with anyone and your responses will be confidential. Please return your questionnaire to Angela Grange in the stamped-addressed envelope provided by Monday __/__/__ if possible. Further information is available about this study; please contact Angela Grange (contact details are on Page 11).

Please time how long it takes you to complete this questionnaire

SECTION 1: YOUR CHILD'S GENERAL HEALTH

1.1 At the moment, how would you describe your child's health?

□ Excellent □ Very good □ Good □ Fair □ Poor

1.2 Compare your child's health now with their health when they were well before (the last) PICU admission by choosing <u>one</u> of the responses below:

My child's health is ***much better now*** than before PICU admission □

My child's health is ***somewhat better now*** than before PICU admission □

My child's health is ***the same now*** as before PICU admission □

My child's health is ***somewhat worse now*** than before PICU admission □

My child's health is ***much worse now*** than before PICU admission □

1.3 Has your child been acutely ill (e.g. poorly, broken bones, accident) in the <u>last two weeks</u>?

□ Yes □ No

If **Yes,** please describe below the symptoms of this illness? *E.g. cough, cold, vomiting, diarrhoea, stomach pain, sore throat, etc.*

……

……

……

……

……

……

……

1.4 Here is list of questions asking you to think about your child's everyday activities in the <u>past two weeks</u>. Indicate how true each statement is of your child in comparison to other healthy children the same age as your child, by circling <u>one response only</u> for each statement, where:

0	=	***much less***	than other children the same age
1	=	***somewhat less***	than other children the same age
2	=	***about the same***	as other children the same age
3	=	***somewhat more***	than other children the same age
4	=	***much more***	than other children the same age
NA	=	***not applicable***	my child is not old enough to do this activity yet

Note: ***If your child was born prematurely, use your child's corrected gestational age, not their chronological age to compare against a healthy child. For example, if your child was born 3 months premature and is now chronologically aged 9 months, compare your child to a child aged 6 months (corrected gestational age).***

	much less than other children	*somewhat less than other*	*about the same as other children*	*somewhat more than other children*	*much more than other children children*	*NA*
My child moves around *(E.g. sits up/rolls over/crawls/takes steps)*	0	1	2	3	4	NA
My child walks or runs	0	1	2	3	4	NA
My child grasps or picks things up	0	1	2	3	4	NA
My child holds & carries things	0	1	2	3	4	NA
My child's height is	0	1	2	3	4	NA
My child's weight is	0	1	2	3	4	NA
My child's vision is	0	1	2	3	4	NA
My child's hearing is	0	1	2	3	4	NA
My child's speech is *(E.g. coos, babbles, talks)*	0	1	2	3	4	NA
My child feels pain	0	1	2	3	4	NA
My child is sad	0	1	2	3	4	NA
My child is happy	0	1	2	3	4	NA
My child is angry	0	1	2	3	4	NA
My child learns	0	1	2	3	4	NA
My child pays attention	0	1	2	3	4	NA
My child understands	0	1	2	3	4	NA
My child plays on his/her own	0	1	2	3	4	NA
My child plays with siblings	0	1	2	3	4	NA
My child plays with friends	0	1	2	3	4	NA
My child cries	0	1	2	3	4	NA
My child laughs	0	1	2	3	4	NA
My child is naughty	0	1	2	3	4	NA
My child behaves him/herself	0	1	2	3	4	NA
My child is tired	0	1	2	3	4	NA
My child is quiet	0	1	2	3	4	NA
My child's tantrums are	0	1	2	3	4	NA

1.5 We are interested in how you saw your child's PICU illness. Indicate how much you feel that the following symptoms were part of the illness that resulted in your child's admission to PICU:

	All of the time	*Frequently*	*Occasionally*	*Never*
Pain or discomfort.....	☐	☐	☐	☐
Feeling sick or vomiting.....	☐	☐	☐	☐
Breathlessness.....	☐	☐	☐	☐
Loss of appetite.....	☐	☐	☐	☐
Lack of energy.....	☐	☐	☐	☐
Sore or sticky eyes.....	☐	☐	☐	☐
Wheeziness	☐	☐	☐	☐
Cough.....	☐	☐	☐	☐
Diarrhoea.....	☐	☐	☐	☐
Sleep difficulties.....	☐	☐	☐	☐
Loss of strength.....	☐	☐	☐	☐
Temperature or fever.....	☐	☐	☐	☐
Runny nose or snuffles.....	☐	☐	☐	☐

1.6 We are interested in your own personal views (not those of your doctor or family) as to how you saw the illness that resulted in your child's admission to PICU. Indicate how much you agree or disagree with the following statements about your child's PICU illness.

At the time of the PICU illness, I thought that:

	Strongly agree	*Agree*	*Neither agree nor disagree*	*Disagree*	*Strongly disagree*
A germ or virus caused my child's PICU illness.....	☐	☐	☐	☐	☐
Diet played a major role in causing my child's PICU illness..... ...	☐	☐	☐	☐	☐
Pollution of the environment caused my child's PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness was hereditary – it runs in my family.....	☐	☐	☐	☐	☐
It was just by chance that my child became ill.....	☐	☐	☐	☐	☐
Stress was a major factor in causing my child's PICU illness..... ...	☐	☐	☐	☐	☐
My child's PICU illness was largely due to his/her own behaviour.....	☐	☐	☐	☐	☐
Other people played a large role in causing my child's PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness was caused by poor medical care in the past.....	☐	☐	☐	☐	☐
My child's state of mind played a major part in causing his/her PICU illness.....	☐	☐	☐	☐	☐
My child's PICU illness would last a short time..... ...	☐	☐	☐	☐	☐
My child's PICU illness was likely to be permanent rather than temporary..... ...	☐	☐	☐	☐	☐
My child's PICU illness would last for a long time.....	☐	☐	☐	☐	☐

At the time of the PICU illness, I thought that:

	Strongly agree	*Agree*	*Neither agree nor disagree*	*Disagree*	*Strongly disagree*
My child's PICU illness was a serious condition.....	☐	☐	☐	☐	☐
My child's PICU illness had major consequences on his/her life..... ...	☐	☐	☐	☐	☐
My child's PICU illness would become easier to live with.....	☐	☐	☐	☐	☐
My child's PICU illness did not have much effect on his/her life..... ...	☐	☐	☐	☐	☐
My child's PICU illness strongly affected the way others saw him/her.....	☐	☐	☐	☐	☐
My child's PICU illness would have serious economic and financial consequences.....	☐	☐	☐	☐	☐
My child's PICU illness strongly affected the way I saw him/her as a person.	☐	☐	☐	☐	☐
My child's PICU illness would improve with time.....	☐	☐	☐	☐	☐
There was a lot which my child could do to control his/her symptoms.....	☐	☐	☐	☐	☐
There was very little that could be done to improve my child's PICU illness.....	☐	☐	☐	☐	☐
Treatment would be effective in curing my child's PICU illness.....	☐	☐	☐	☐	☐
My child's recovery from PICU illness was largely dependent on chance or fate...	☐	☐	☐	☐	☐
What my child did could determine whether his/her PICU illness got better or worse.. ...	☐	☐	☐	☐	☐

1.7 How often during the <u>past two weeks</u> has your child's health:

	Very often	*Quite often*	*Occasionally*	*Almost never*	*Never*
Interrupted everyday family activities *(E.g. mealtimes)*	☐	☐	☐	☐	☐
Stopped you from going out at short notice	☐	☐	☐	☐	☐
Caused tension or arguments in your home	☐	☐	☐	☐	☐
Caused you to cancel or change plans at the last minute at home or work	☐	☐	☐	☐	☐
Stopped you from going on a family outing	☐	☐	☐	☐	☐

1.8 During the <u>past two weeks</u> how much have you been worried about your child's physical health?

☐	☐	☐	☐	☐
Not at all	*A little bit*	*Some*	*Quite a bit*	*A lot*

1.9 During the <u>past two weeks</u> how much have you been worried about your child's emotional state?

☐ *Not at all* ☐ *A little bit* ☐ *Some* ☐ *Quite a bit* ☐ *A lot*

1.10 During the <u>past two weeks</u> how much have you been worried about your child's behaviour?

☐ *Not at all* ☐ *A little bit* ☐ *Some* ☐ *Quite a bit* ☐ *A lot*

1.11 During the <u>past two weeks</u> how much have you been worried about your child's learning abilities?

☐ *Not at all* ☐ *A little bit* ☐ *Some* ☐ *Quite a bit* ☐ *A lot*

1.12 Describe how you <u>feel right now</u> about <u>your child's health and wellbeing</u> by placing a circle around the most appropriate number to the right of each statement.

	Not at all	*Somewhat*	*Moderately*	*Very much*
I feel calm.....	1	2	3	4
I am tense.....	1	2	3	4
I feel upset.....	1	2	3	4
I am relaxed.....	1	2	3	4
I feel content.....	1	2	3	4
I am worried.....	1	2	3	4

SECTION 2: YOUR CHILD'S RESPIRATORY HEALTH

2.1 How would you describe your child's breathing now?

☐ Excellent ☐ Very good ☐ Good ☐ Fair ☐ Poor

2.2 Compare your child's level of breathing now with their level of breathing when they were well before (the last) PICU admission by choosing <u>one</u> of the responses below:

My child's breathing is ***much better now*** than before PICU admission ☐

My child's breathing is ***somewhat better now*** than before PICU admission ☐

My child's breathing is ***the same now*** as before PICU admission ☐

My child's breathing is ***somewhat worse now*** than before PICU admission ☐

My child's breathing is ***much worse now*** than before PICU admission ☐

2.3 a) i) How often has your child experienced a day-time cough in the <u>past two weeks</u>? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

ii) How serious do you think this day-time cough was in the <u>past two weeks</u>? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle 0 = not applicable)*

Very serious — *Somewhat serious* — *Moderately serious* — *A little serious* — *Not at all serious* — *Not applicable*

5 — 4 — 3 — 2 — 1 — 0

b) i) How often has your child experienced a night-time cough in the past two weeks? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

ii) How serious do you think this night-time cough was in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle* 0 *= not applicable)*

Very serious — *Somewhat serious* — *Moderately serious* — *A little serious* — *Not at all serious* — *Not applicable*

5 4 3 2 1 0

c) i) How often has your child experienced making noisy sounds on breathing in (*a stridor*) in the past two weeks? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

ii) How serious do you think these noisy sounds on breathing in (*a stridor*) were in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle* 0 *= not applicable)*

Very serious — *Somewhat serious* — *Moderately serious* — *A little serious* — *Not at all serious* — *Not applicable*

5 4 3 2 1 0

d) i) How often has your child experienced making noisy sounds on breathing out (*a wheeze*) in the past two weeks? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

ii) How serious do you think these noisy sounds on breathing out (a wheeze) were in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle* 0 *= not applicable)*

Very serious — *Somewhat serious* — *Moderately serious* — *A little serious* — *Not at all serious* — *Not applicable*

5 4 3 2 1 0

e) i) How often has your child experienced breathing at a faster rate than normal in the past two weeks? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

ii) How serious do you think this breathing at a faster rate than normal was in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle* 0 *= not applicable)*

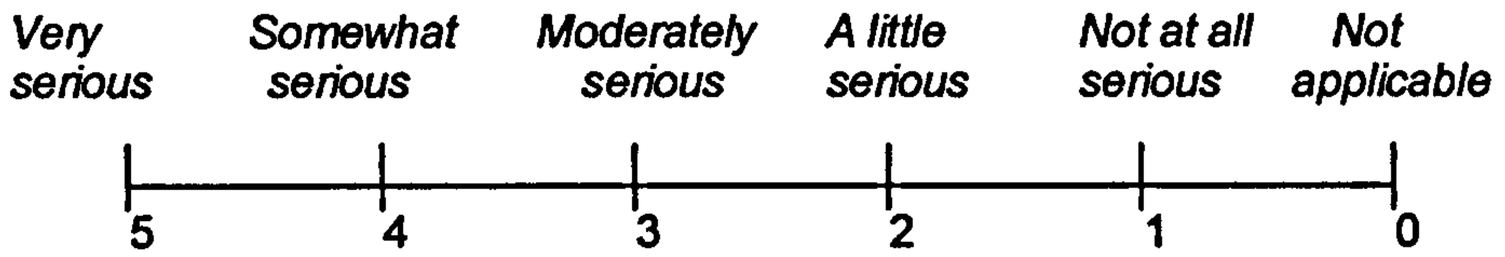
Very serious — *Somewhat serious* — *Moderately serious* — *A little serious* — *Not at all serious* — *Not applicable*

5 4 3 2 1 0

f) i) How often has your child experienced breathing at a slower rate than normal in the past two weeks? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

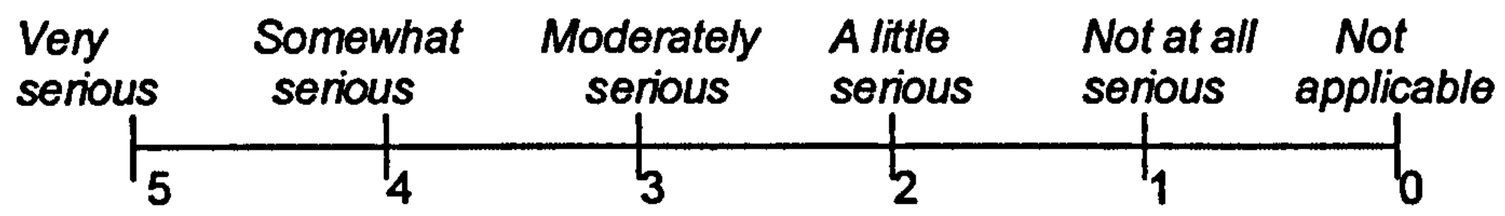
ii) How serious do you think this breathing at a slower rate than normal was in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle 0 = not applicable)*



g) i) How often has your child experienced breath-holding episodes (apnoeas) in the past two weeks? *(tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

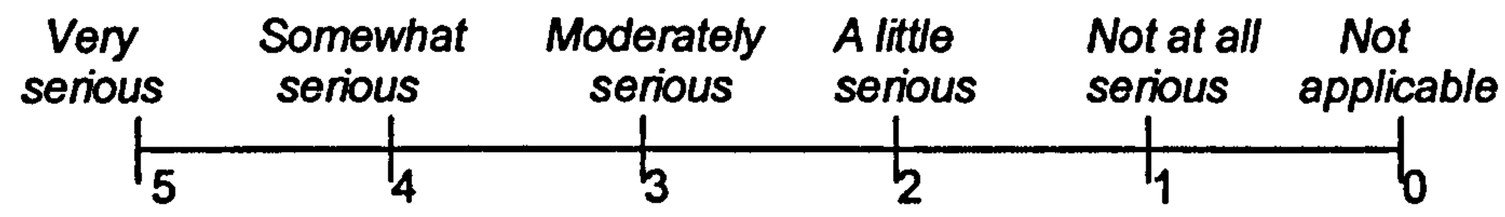
ii) How serious do you think these breath-holding episodes were in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle 0 = not applicable)*



h) i) How often has your child experienced breathlessness (being out of breath) when still or inactive and resting in the past two weeks? *(tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

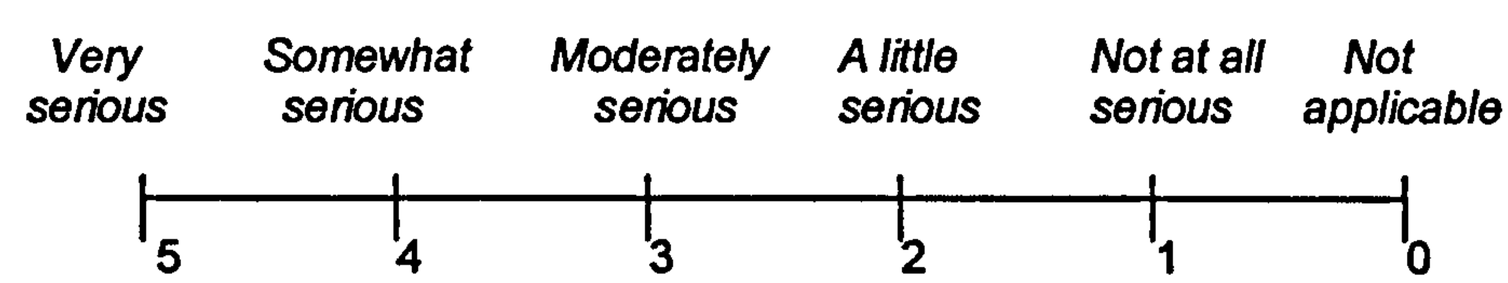
ii) How serious do you think this breathlessness when still or inactive and resting was in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle 0 = not applicable)*



i) i) How often has your child experienced breathlessness (being out of breath) when active, e.g. playing, crawling, walking, running, in the past two weeks? *(Please tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

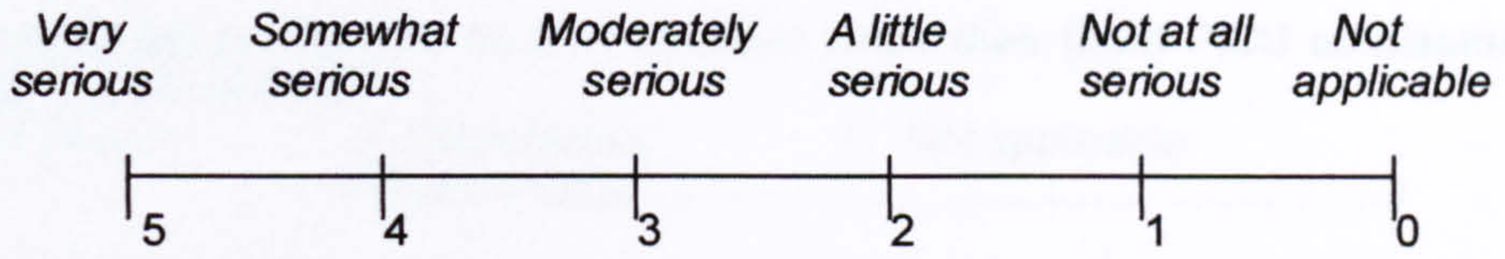
ii) How serious do you think this breathlessness when active was in the past two weeks? *(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle 0 = not applicable)*



j) i) How often has your child had a runny nose or snuffles in the past two weeks? *(tick ONE box only)*

☐ *Every day* ☐ *Every 2-3 days* ☐ *Every week* ☐ *Every 2 weeks* ☐ *Not at all*

ii) How serious do you think this runny nose or snuffles were in the past two weeks?
(Please circle ONE of the numbers on the scale below. If your child did not have this symptom circle 0 = not applicable)



2.4 Has your child been limited in any of the following everyday activities because of breathing (respiratory) problems in the past two weeks (*Please tick ONE box per activity)?*

		Not limited at all	*Sometimes limited*	*Often limited*	*Always limited*
a)	Eating/feeding	☐	☐	☐	☐
b)	Drinking	☐	☐	☐	☐
c)	Moving around *(E.g. sitting/rolling over/crawling/walking)*	☐	☐	☐	☐
d)	Sleeping	☐	☐	☐	☐
e)	Playing	☐	☐	☐	☐
f)	Communicating *(E.g. cooing/babbling/talking)*	☐	☐	☐	☐

SECTION 3: FACTS ABOUT YOUR CHILD

3.1 Is your child?

☐ Male ☐ Female

3.2a) Was your child born?

☐ First (eldest) ☐ Second ☐ Third ☐ Fourth ☐ Other* (**Please specify*.....)

b) Is your child a ☐ Singleton ☐ Twin ☐ Triplet ☐ Other* (**Please specify*.....)

3.3 Was your child born prematurely?

☐ Yes ☐ No

If **Yes,** at what age (in weeks) was your child born *(e.g. 26 weeks)*? ☐☐ weeks

3.4 What is your child's date of birth?

☐☐ / ☐☐ / ☐☐
Day Month Year

3.5 How many times has your child been admitted to hospital as an in-patient before their (last) PICU admission? *(Do not count the time your child was in hospital while being born)*

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.6 If your child was admitted to hospital as an in-patient before their (last) PICU admission, what was the main reason for this?

☐ Breathing (respiratory) problem ☐ Born prematurely ☐ Other illness *(Please specify.....)* ☐ Not applicable

3.7 How many times has your child been admitted to hospital as an in-patient since their (last) PICU admission?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.8 If your child was admitted to hospital as an in-patient since their (last) PICU admission, what was the main reason for this?

☐ Breathing (respiratory) problem ☐ Other illness *(Please specify.....)* ☐ Not applicable

3.9 Has your child ever had oxygen therapy for a breathing problem before their PICU admission?

☐ Yes ☐ No ☐ Don't know

3.10 Has your child ever had steroids to treat a breathing problem before their PICU admission?

☐ Yes ☐ No ☐ Don't know

3.11 Has your child ever been artificially ventilated (i.e. put on a ventilator or breathing machine) for a breathing problem before their PICU admission?

☐ Yes ☐ No ☐ Don't know

If Yes, a) How long were they artificially ventilated for?
..... days/weeks* (*please delete as appropriate)

b) How long has it been since your child was last artificially ventilated?
..... weeks/months* (*please delete as appropriate)

3.12 Does your child currently receive oxygen therapy?

☐ Yes ☐ No

If Yes, a) How often is the oxygen given?
☐ Continuously ☐ Intermittently

b) How much oxygen does your child get in litres/minute during the day/night?
☐.☐/litres/minute in the day ☐.☐/litres/minute at night

3.13 In the <u>last month</u>, how many times have you visited/called out your GP for any reason related to your <u>child's health</u>?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.14 In the <u>last month</u>, how many times have you visited/called out your GP because of a problem with your <u>child's breathing</u>?

☐ None ☐ One ☐ Two ☐ Three ☐ Four ☐ Five ☐ More than five

3.15 Has a nurse, doctor or other health care professional ever told you that your child has any of the following chest or lung conditions? *(Please tick ONE box per line)*

	Yes	*No*	*Don't know*
Asthma.....	☐	☐	☐
Chronic respiratory, lung or breathing problem *not asthma*.	☐	☐	☐
Cystic fibrosis.....	☐	☐	☐

3.16 Has a nurse, doctor or other health care professional ever told you that your child has any of the following conditions? *(Please tick ONE box per line)*

	Yes	*No*	*Don't know*
Anxiety problems.....	☐	☐	☐
Behavioural problems.....	☐	☐	☐
Cancer.....	☐	☐	☐

	Yes	No	Don't know
Cerebral palsy	☐	☐	☐
Chest abnormality (e.g. pigeon chest)	☐	☐	☐
Chronic allergies or sinus trouble	☐	☐	☐
Chronic orthopaedic, bone or joint problems	☐	☐	☐
Developmental delay	☐	☐	☐
Digestive (eating and drinking or gut) problems, e.g. reflux	☐	☐	☐
Down's syndrome	☐	☐	☐
Epilepsy (seizure disorder) or fits	☐	☐	☐
Hearing impairment or deafness	☐	☐	☐
Heart problems	☐	☐	☐
Immune problems	☐	☐	☐
Kidney/bladder problems	☐	☐	☐
Learning problems	☐	☐	☐
Liver problems	☐	☐	☐
Neurological (brain/nervous system) problems	☐	☐	☐
Sleep disturbance	☐	☐	☐
Speech or language problems	☐	☐	☐
Vision problems	☐	☐	☐
Any other health problem*	☐	☐	☐

(**Please describe*.....)

SECTION 4: FACTS ABOUT YOU & YOUR FAMILY

4.1 Are you?

☐ Male ☐ Female

4.2 What is your date of birth?

☐☐ / ☐☐ / ☐☐
Day Month Year

4.3 Which one of the following best describes your ethnic origin?

☐ White
☐ Black – Caribbean
☐ Black – African
☐ Black – other *(Please specify* .....*)*
☐ Indian
☐ Pakistani
☐ Bangladeshi
☐ Chinese
☐ Other *(Please specify* .....*)*

4.4 Which one of the following best describes your marital status?

☐ Married ☐ Living together as a couple ☐ Widowed ☐ Divorced or separated ☐ Single

4.5 Which one of the following best describes your relationship to your child?

☐ Biological parent ☐ Step-parent ☐ Adoptive parent ☐ Foster parent ☐ Other (*Please state*.....)

4.6 Has your work status changed since your child was discharged from PICU?

☐ Yes ☐ No

'Original in colour'

If **Yes**, please state how it has changed? *(E.g. I now work part-time/I now work full-time/I now do not work because of my child's health problems)*.....
.....
.....
.....

4.7 What is the highest grade of educational qualification that you have completed?

☐	☐	☐	☐	☐
No formal education	GCSE or O level	A level	Professional qualification	Degree or or higher

4.8 Do you or anyone in your family smoke in your home?

☐ Yes ☐ No

4.9 Do you have any pets in your home?

☐ Yes ☐ No

If **Yes**, please specify type:.....

4.10 What is today's date?

☐☐ / ☐☐ / ☐☐
Day Month Year

SECTION 5: YOUR VIEWS ABOUT THIS QUESTIONNAIRE

5.1 How many minutes (approximately) did it take you to complete this questionnaire?

☐☐ minutes

5.2 How easy did you find it to complete this questionnaire?

☐	☐	☐	☐	☐
Very easy	Quite easy	Neither easy nor difficult	Quite difficult	Difficult

If you have any comments about this questionnaire or your child's health and care in general, write them below:

COMMENTS:

Please return your questionnaire to Angela Grange
C/o Ward 2
Jubilee Wing
Leeds General Infirmary
Great George Street
Leeds LS1 3EX
Tel: 0113 392 3220
Thank you for your participation

Appendix XI - Syntax in SPSS & formulae to calculate PICQoL scores

Positive milestones score (Qu 1.4)

```
COMPUTE summile = 5*(holds_1+grasps_1) +
4*(moves_1+walks_1+speech_1+learns_1+under_1+sibpl_1+friepl_1+naught_1)
+ (3*(vision_1) + 2*(height_1+weight_1+hears_1+attent_1+ownpl_1+laughs_1).
EXECUTE.
COMPUTE milescor = (summile-0/228-0)*100.
EXECUTE.
```

Maximum positive milestones score (Qu 1.4)

5 x (4 + 4) ADD 4 x (4 + 4 + 4 + 4 + 4 + 4 + 4 + 4) ADD 3 x (4) ADD 2 X (4 + 4 + 4 + 4+ 4 + 4)
= 40 + 128 + 12 + 48
= 228

Minimum milestones score (Qu 1.4)

5 x (0 + 0) ADD 4 x (0 + 0 + 0 + 0 + 0 + 0 + 0 + 0) ADD 3 x (0) ADD 2 x (0 + 0 + 0 + 0 + 0 + 0)
= 0

$$\text{Transformed milestones score} = \frac{(\text{actual score} - 0)}{228} \times 100$$

(100 = best score and 0 = worst score)

Negative emotions score Qu 1.4

```
COMPUTE sumemot = 4*(tantru_1+angry_1+cries_1) + 3*(sad_1-happy_1+naught_1) +
2*(pain_1-behave_1).
EXECUTE.
COMPUTE emotscor = (80-sumemot)/80-(-20))*100.
EXECUTE.
```

Maximum negative emotions score Qu 1.4

4 x (4 + 4 + 4) ADD 3 x (4 – 0 + 4) ADD 2 x (4 – 0)
= 48 + 24 + 8
= 80

Minimum negative emotions score Qu 1.4

4 x (0 + 0 + 0) ADD 3 x (0 – 4 + 0) ADD 2 x (0 – 4)
= 0 + (-12) + (-8)
= -20

$$\text{The transformed negative emotions score} = \frac{80 - \text{actual score}}{80 - (-20)} \times 100$$

(100 = best score and 0= worst score)

Impact on family score (Qu. 1.7)

```
COMPUTE val1_7 = NVALID(family,goingout, tension, plans,outing) .
EXECUTE .

RECODE
  family goingout tension plans outing
  (1=1) (2=2) (3=3) (4=4) (5=5) (MISSING=0) INTO familyn goingoun
  tensionn plansn outingn .
EXECUTE .

IF (val1_7 >= 3) impact = (SUM(familyn,goingoun,tensionn, plansn, outingn)) / val1_7 .
VARIABLE LABELS impact 'Impact on family' .
EXECUTE .

COMPUTE  score1_7 = ((impact - 1) / 4) * 100 .
EXECUTE .
```

Parental worries about health score (Qu. 1.8-1.11)

```
COMPUTE val1_8 = NVALID (physical, emotions, behaviou, learning)

RECODE physical emotions behaviou learning
(1=5) (2=4) (3=3) (4=2) (5=1) (MISSING=0) INTO physican emotionn behavion learninn.
EXECUTE.

IF (val1_8 >=2) worries = (SUM (physican, emotionn, behavion, learninn))/ val1_8.
VARIABLE LABELS worries 'parental worries about child health'.
EXECUTE.

COMPUTE score1_8 = ((worries-1) / 4) * 100.
EXECUTE.
```

Respiratory limitations score Qu 2.4

```
COMPUTE  val2_4 = NVALID (leating, ldrinkin,lmoving, lsleepin, lplaying, lcommun).

RECODE leating ldrinkin lmoving lsleepin lplaying lcommun
(1=4) (2=3) (3=2) (4=1) (MISSING=0) INTO leatingr ldrinkr lmovingr lsleepr lplayr
lcommunr.
EXECUTE.

IF (val2_4 >=3) resplim = (SUM (leatingr, ldrinkr, lmovingr, lsleepr, lplayr, lcommunr)) /
val2_4.
VARIABLE LABELS resplim 'limitations owing to breathing problems'.
EXECUTE.
COMPUTE score2_4 = ((resplim -1) / 3) * 100.
EXECUTE.
```

Recoding of Qu 2.3 items (day-time cough illustrated)

```
COMPUTE fdtcougr = fdtcough .
COMPUTE sdtcougr = sdtcough .
EXECUTE .
RECODE
  fdtcougr sdtcougr  (6=SYSMIS)  .
EXECUTE .

IF (fdtcough = 5) sdtcougr = 0 .
EXECUTE .
IF ((sdtcough = 0) & MISSING(fdtcough)) fdtcougr = 5 .
EXECUTE .
DO IF (fdtcough  <= 4) .
RECODE
  sdtcougr  (0=SYSMIS)  .
END IF .
EXECUTE .
```

Maximum respiratory symptom score

5 x (5 + 5 + 5 + 5 + 5 + 5 + 5) ADD 4 x (4 + 4 + 4 + 4 + 4 + 4 + 4) ADD 3 x (5 + 5) ADD 2 x (4 + 4)
= 175 + 112 + 30 + 16
= 333

Minimum respiratory symptom score

5 x (0 + 0 + 0 + 0 + 0 + 0 + 0) ADD 4 x (0 + 0 + 0 + 0 + 0 + 0 + 0) ADD 3 x (0 + 0) ADD 2 X (0 + 0)
= 0 + 0 + 0 + 0
= 0

$$\text{Transformed respiratory score} = \frac{333 - \text{actual score}}{333 - 0} \times 100$$

Respiratory symptom score

```
COMPUTE sumresp=5*(sdtcou_1+sntcou_1+sstrid_1+swheez_1+sfast_1+sstill_1
+sactiv_1) + 4*(fdtcou_1+fntcou_1+fstrid_1+fwheez_1+ffast_1+fstill_1+factiv_1) +
3*(ssnuff_1+sslowe_1) + 2*(fsnuff_1+fslowe_1).
EXECUTE.
COMPUTE respscor = ((333-sumresp)/333)*100.
EXECUTE.
```

Appendix XII – PCA of daily activity items (Question 1.4) - merged data

PCA (1-factor solution) of Qu. 1.4 merged Phase II/III data

Total Variance Explained

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	7.045	27.097	27.097	7.045	27.097	27.097
2	3.020	11.614	38.711			
3	1.919	7.381	46.093			
4	1.439	5.533	51.626			
5	1.389	5.342	56.968			
6	1.269	4.880	61.848			
7	1.123	4.320	66.168			
8	1.034	3.978	70.146			
9	.898	3.453	73.599			
10	.828	3.185	76.784			
11	.799	3.072	79.856			
12	.647	2.489	82.345			
13	.555	2.133	84.478			
14	.550	2.114	86.592			
15	.534	2.054	88.646			
16	.456	1.752	90.398			
17	.422	1.622	92.020			
18	.341	1.313	93.332			
19	.331	1.272	94.604			
20	.284	1.091	95.695			
21	.262	1.009	96.704			
22	.247	.949	97.653			
23	.185	.711	98.365			
24	.173	.666	99.030			
25	.153	.587	99.617			
26	.100	.383	100.000			

Extraction Method: Principal Component Analysis.

Component Matrix[a]

	Component
	1
Learns recoded (medians)	.792
Understands recoded (medians)	.785
Speech recoded (medians)	.778
Holds recoded (medians)	.747
Plays with friends recoded (medians)	.714
Moves recoded (medians)	.714
Grasps recoded (medians)	.709
Walks recoded (medians)	.664
Plays with siblings recoded (medians)	.578
Pays attention recoded (medians)	.547
Happy recoded (medians)	.530
Laughs recoded (medians)	.503
Height recoded (medians)	.486
Vision recoded (medians)	.474
Sad recoded (medians)	-.454
Naughty recoded (medians)	.429
Plays on own recoded (medians)	.398
Weight recoded (medians)	.365
Hearing recoded (medians)	.363
Quiet recoded (medians)	
Cries recoded (medians)	
Tired recoded (medians)	
Behaves recoded (medians)	
Tantrums recoded (medians)	
Angry recoded (medians)	
Pain recoded (medians)	

Extraction Method: Principal Component Analysis.

a. 1 components extracted.

Note: Factor loading scores less than 0.3 are shown as blank in this table

PCA (3-factor solution) of Qu. 1.4 merged Phase II/III data

Total Variance Explained

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	7.045	27.097	27.097	7.045	27.097	27.097	6.032	23.200	23.200
2	3.020	11.614	38.711	3.020	11.614	38.711	3.063	11.780	34.979
3	1.919	7.381	46.093	1.919	7.381	46.093	2.889	11.113	46.093
4	1.439	5.533	51.626						
5	1.389	5.342	56.968						
6	1.269	4.880	61.848						
7	1.123	4.320	66.168						
8	1.034	3.978	70.146						
9	.898	3.453	73.599						
10	.828	3.185	76.784						
11	.799	3.072	79.856						
12	.647	2.489	82.345						
13	.555	2.133	84.478						
14	.550	2.114	86.592						
15	.534	2.054	88.646						
16	.456	1.752	90.398						
17	.422	1.622	92.020						
18	.341	1.313	93.332						
19	.331	1.272	94.604						
20	.284	1.091	95.695						
21	.262	1.009	96.704						
22	.247	.949	97.653						
23	.185	.711	98.365						
24	.173	.666	99.030						
25	.153	.587	99.617						
26	.100	.383	100.000						

Extraction Method: Principal Component Analysis.

Rotated Component Matrix[a]

	Component		
	1	2	3
Understands recoded (medians)	.757		
Learns recoded (medians)	.751		
Plays with friends recoded (medians)	.733		
Speech recoded (medians)	.714		
Holds recoded (medians)	.701		.339
Pays attention recoded (medians)	.662		
Grasps recoded (medians)	.637		.367
Plays with siblings recoded (medians)	.593		
Moves recoded (medians)	.572		.494
Vision recoded (medians)	.528		
Walks recoded (medians)	.524		.469
Hearing recoded (medians)	.517		
Plays on own recoded (medians)	.455		
Cries recoded (medians)		.735	
Angry recoded (medians)		.699	
Tantrums recoded (medians)		.673	
Naughty recoded (medians)	.398	.538	.301
Sad recoded (medians)	-.357	.513	
Happy recoded (medians)	.443	-.509	
Behaves recoded (medians)	.314	-.392	-.365
Pain recoded (medians)		.344	
Quiet recoded (medians)			-.701
Weight recoded (medians)			.557
Height recoded (medians)			.524
Laughs recoded (medians)	.310		.450
Tired recoded (medians)			-.381

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 8 iterations.

Appendix XIII - PCA of respiratory items (Question 2.3) – Phases II and III data

PCA (2-factor solution) varimax rotation Qu 2.3 Phase II data

Communalities

	Initial	Extraction
MEDIAN(FDTCOURR,ALL)	1.000	.600
MEDIAN(SDTCOUGR,ALL)	1.000	.799
MEDIAN (FNTCOURR, ALL)	1.000	.590
MEDIAN(SNTCOUGR,ALL)	1.000	.786
MEDIAN(FSTRIRR,ALL)	1.000	.447
MEDIAN(SSTRIDRR,ALL)	1.000	.702
MEDIAN(FWHEEZRR,ALL)	1.000	.551
MEDIAN(SWHEEZER,ALL)	1.000	.730
MEDIAN(FFASTRR,ALL)	1.000	.560
MEDIAN(SFASTERR,ALL)	1.000	.662
MEDIAN(FSLOWRR,ALL)	1.000	.284
MEDIAN(SSLOWERR,ALL)	1.000	.308
MEDIAN(FSTILLRR,ALL)	1.000	.400
MEDIAN(SSTILLR,ALL)	1.000	.474
MEDIAN(FACTIVRR,ALL)	1.000	.491
MEDIAN(SACTIVER,ALL)	1.000	.612
MEDIAN(FSNUFFRR,ALL)	1.000	.545
MEDIAN(SSNUFFLR,ALL)	1.000	.653

Extraction Method: Principal Component Analysis.

Total Variance Explained

	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
Component	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	8.061	44.784	44.784	8.061	44.784	44.784	6.458	35.875	35.875
2	2.133	11.849	56.633	2.133	11.849	56.633	3.736	20.758	56.633
3	1.978	10.990	67.624						
4	1.377	7.651	75.275						
5	.914	5.080	80.355						
6	.836	4.647	85.002						
7	.790	4.391	89.394						
8	.641	3.563	92.957						
9	.307	1.705	94.662						
10	.230	1.275	95.937						
11	.172	.956	96.893						
12	.166	.921	97.813						
13	.134	.747	98.560						
14	.087	.483	99.043						
15	.068	.378	99.422						
16	.047	.263	99.685						
17	.042	.231	99.916						
18	.015	8.39E-02	100.000						

Extraction Method: Principal Component Analysis.

Note: 8.39E-02 = 0.0839

Component Matrix[a]

	Component	
	1	2
MEDIAN(SWHEEZER,ALL)	.827	
MEDIAN(SSTRIDRR,ALL)	.825	
MEDIAN(SFASTERR,ALL)	.813	
MEDIAN(SNTCOUGR,ALL)	.809	.362
MEDIAN(SDTCOUGR,ALL)	.809	.380
MEDIAN(FFASTRR,ALL)	.748	
MEDIAN(FDTCOURR,ALL)	.706	.319
MEDIAN(SACTIVER,ALL)	.702	-.345
MEDIAN(SSTILLR,ALL)	.682	
MEDIAN (FNTCOURR, ALL)	.682	.354
MEDIAN(FWHEEZRR,ALL)	.669	-.322
MEDIAN(FSTRIRR,ALL)	.634	
MEDIAN(FSTILLRR,ALL)	.593	
MEDIAN(FACTIVRR,ALL)	.588	-.382
MEDIAN(SSLOWERR,ALL)	.535	
MEDIAN(FSLOWRR,ALL)	.484	
MEDIAN(FSNUFFRR,ALL)		.723
MEDIAN(SSNUFFLR,ALL)	.394	.706

Extraction Method: Principal Component Analysis.

a. 2 components extracted.

Rotated Component Matrix[a]

	Component	
	1	2
MEDIAN(SWHEEZER,ALL)	.818	
MEDIAN(SSTRIDRR,ALL)	.782	.303
MEDIAN(SACTIVER,ALL)	.779	
MEDIAN(FWHEEZRR,ALL)	.739	
MEDIAN(FACTIVRR,ALL)	.701	
MEDIAN(SFASTERR,ALL)	.678	.449
MEDIAN(FSTRIRR,ALL)	.652	
MEDIAN(SSTILLR,ALL)	.629	
MEDIAN(FFASTRR,ALL)	.628	.407
MEDIAN(FSTILLRR,ALL)	.620	
MEDIAN(SSLOWERR,ALL)	.535	
MEDIAN(FSLOWRR,ALL)	.529	
MEDIAN(SSNUFFLR,ALL)		.808
MEDIAN(SDTCOUGR,ALL)	.494	.745
MEDIAN(SNTCOUGR,ALL)	.503	.730
MEDIAN(FSNUFFRR,ALL)		.696
MEDIAN (FNTCOURR, ALL)	.398	.657
MEDIAN(FDTCOURR,ALL)	.437	.640

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 3 iterations.

Component Transformation Matrix

Component	1	2
1	.854	.520
2	-.520	.854

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

Factor 1 = 'miscellaneous symptom' factor
Factor 2 = 'cough and snuffles' factor

PCA (2-factor solution) varimax rotation Phase III

Communalities

	Initial	Extraction
MEDIAN(FDTCOURR,ALL)	1.000	.480
MEDIAN(SDTCOUGR,ALL)	1.000	.709
MEDIAN (FNTCOUGR, ALL)	1.000	.449
MEDIAN(SNTCOUGR,ALL)	1.000	.634
MEDIAN(FSTRIRR,ALL)	1.000	.519
MEDIAN(SSTRIDRR,ALL)	1.000	.684
MEDIAN(FWHEEZRR,ALL)	1.000	.541
MEDIAN(SWHEEZER,ALL)	1.000	.729
MEDIAN(FFASTRR,ALL)	1.000	.583
MEDIAN(SFASTERR,ALL)	1.000	.754
MEDIAN(FSLOWRR,ALL)	1.000	.513
MEDIAN(SSLOWERR,ALL)	1.000	.381
MEDIAN(FAPNOERR,ALL)	1.000	.789
MEDIAN(SAPNOEAR,ALL)	1.000	.708
MEDIAN(FSTILLRR,ALL)	1.000	.553
MEDIAN(SSTILLR,ALL)	1.000	.535
MEDIAN(FACTIVRR,ALL)	1.000	.483
MEDIAN(SACTIVER,ALL)	1.000	.605
MEDIAN(FSNUFFRR,ALL)	1.000	.215
MEDIAN(SSNUFFLR,ALL)	1.000	.385

Extraction Method: Principal Component Analysis.

Total Variance Explained

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	9.081	45.403	45.403	9.081	45.403	45.403	8.355	41.773	41.773
2	2.171	10.854	56.256	2.171	10.854	56.256	2.897	14.484	56.256
3	1.727	8.636	64.892						
4	1.330	6.649	71.542						
5	1.112	5.558	77.100						
6	.942	4.709	81.809						
7	.792	3.961	85.770						
8	.618	3.089	88.859						
9	.391	1.956	90.815						
10	.374	1.870	92.684						
11	.332	1.660	94.344						
12	.254	1.272	95.617						
13	.225	1.123	96.740						
14	.158	.788	97.528						
15	.148	.738	98.267						
16	.112	.560	98.827						
17	.081	.404	99.231						
18	.073	.365	99.596						
19	.045	.226	99.822						
20	.036	.178	100.000						

Extraction Method: Principal Component Analysis.

Component Matrix[a]

	Component	
	1	2
MEDIAN(SWHEEZER,ALL)	.848	
MEDIAN(SFASTERR,ALL)	.844	
MEDIAN(SDTCOUGR,ALL)	.840	
MEDIAN(SSTRIDRR,ALL)	.817	
MEDIAN(SNTCOUGR,ALL)	.795	
MEDIAN(FFASTRR,ALL)	.759	
MEDIAN(SACTIVER,ALL)	.757	
MEDIAN(FWHEEZRR,ALL)	.735	
MEDIAN(FSTRIRR,ALL)	.721	
MEDIAN(FACTIVRR,ALL)	.695	
MEDIAN(FDTCOURR,ALL)	.692	
MEDIAN(FSTILLRR,ALL)	.675	-.313
MEDIAN (FNTCOUGR, ALL)	.669	
MEDIAN(SSTILLR,ALL)	.667	-.301
MEDIAN(SSNUFFLR,ALL)	.593	
MEDIAN(SSLOWERR,ALL)	.477	.391
MEDIAN(FSNUFFRR,ALL)	.461	
MEDIAN(FAPNOERR,ALL)	.305	.835
MEDIAN(SAPNOEAR,ALL)	.323	.777
MEDIAN(FSLOWRR,ALL)	.369	.614

Extraction Method: Principal Component Analysis.

a. 2 components extracted.

Rotated Component Matrix[a]

	Component	
	1	2
MEDIAN(SFASTERR,ALL)	.864	
MEDIAN(SWHEEZER,ALL)	.834	
MEDIAN(SSTRIDRR,ALL)	.815	
MEDIAN(SDTCOUGR,ALL)	.814	
MEDIAN(SACTIVER,ALL)	.774	
MEDIAN(SNTCOUGR,ALL)	.767	
MEDIAN(FFASTRR,ALL)	.745	
MEDIAN(FSTILLRR,ALL)	.740	
MEDIAN(SSTILLR,ALL)	.728	
MEDIAN(FWHEEZRR,ALL)	.703	
MEDIAN(FSTRIRR,ALL)	.687	
MEDIAN(FACTIVRR,ALL)	.653	
MEDIAN(FDTCOURR,ALL)	.650	
MEDIAN (FNTCOUGR, ALL)	.625	
MEDIAN(SSNUFFLR,ALL)	.502	.365
MEDIAN(FSNUFFRR,ALL)	.418	
MEDIAN(FAPNOERR,ALL)		.888
MEDIAN(SAPNOEAR,ALL)		.840
MEDIAN(FSLOWRR,ALL)		.701
MEDIAN(SSLOWERR,ALL)	.325	.525

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 3 iterations.

Component Transformation Matrix

Component	1	2
1	.946	.324
2	-.324	.946

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

Factor 1 = 'miscellaneous symptom' factor
Factor 2 = 'slow breathing' factor

PCA (2-factor solution) varimax rotation Qu 2.3 merged Phase II/III data

Communalities

	Initial	Extraction
MEDIAN(FDTCOURR,ALL)	1.000	.614
MEDIAN(SDTCOUGR,ALL)	1.000	.753
MEDIAN (FNTCOUGR, ALL)	1.000	.632
MEDIAN(SNTCOUGR,ALL)	1.000	.740
MEDIAN(FSTRIRR,ALL)	1.000	.511
MEDIAN(SSTRIDRR,ALL)	1.000	.718
MEDIAN(FWHEEZRR,ALL)	1.000	.548
MEDIAN(SWHEEZER,ALL)	1.000	.725
MEDIAN(FFASTRR,ALL)	1.000	.588
MEDIAN(SFASTERR,ALL)	1.000	.682
MEDIAN(FSLOWRR,ALL)	1.000	.280
MEDIAN(SSLOWERR,ALL)	1.000	.333
MEDIAN(FAPNOERR,ALL)	1.000	.141
MEDIAN(SAPNOEAR,ALL)	1.000	5.565E-02
MEDIAN(FSTILLRR,ALL)	1.000	.539
MEDIAN(SSTILLR,ALL)	1.000	.619
MEDIAN(FACTIVRR,ALL)	1.000	.523
MEDIAN(SACTIVER,ALL)	1.000	.657
MEDIAN(FSNUFFRR,ALL)	1.000	.456
MEDIAN(SSNUFFLR,ALL)	1.000	.477

Extraction Method: Principal Component Analysis.

Total Variance Explained

	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
Component	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	8.529	42.646	42.646	8.529	42.646	42.646	5.890	29.452	29.452
2	2.062	10.312	52.959	2.062	10.312	52.959	4.701	23.507	52.959
3	1.620	8.099	61.057						
4	1.318	6.591	67.648						
5	1.047	5.234	72.882						
6	.998	4.991	77.873						
7	.886	4.430	82.303						
8	.693	3.464	85.767						
9	.666	3.330	89.097						
10	.611	3.056	92.154						
11	.393	1.964	94.117						
12	.278	1.392	95.509						
13	.237	1.186	96.695						
14	.176	.878	97.573						
15	.155	.777	98.350						
16	.096	.480	98.830						
17	.084	.421	99.251						
18	.061	.304	99.555						
19	.052	.262	99.817						
20	.037	.183	100.000						

Extraction Method: Principal Component Analysis.

Component Matrix[a]

	Component	
	1	2
MEDIAN(SWHEEZER,ALL)	.847	
MEDIAN(SSTRIDRR,ALL)	.834	
MEDIAN(SFASTERR,ALL)	.825	
MEDIAN(SDTCOUGR,ALL)	.806	.321
MEDIAN(SNTCOUGR,ALL)	.786	.349
MEDIAN(FFASTRR,ALL)	.767	
MEDIAN(FWHEEZRR,ALL)	.739	
MEDIAN(SACTIVER,ALL)	.737	-.337
MEDIAN(SSTILLR,ALL)	.712	-.335
MEDIAN(FSTRIRR,ALL)	.712	
MEDIAN(FDTCOURR,ALL)	.676	.396
MEDIAN(FACTIVRR,ALL)	.643	-.331
MEDIAN (FNTCOUGR, ALL)	.632	.483
MEDIAN(FSTILLRR,ALL)	.631	-.375
MEDIAN(SSNUFFLR,ALL)	.494	.483
MEDIAN(SSLOWERR,ALL)	.485	-.313
MEDIAN(FSLOWRR,ALL)	.427	-.313
MEDIAN(SAPNOEAR,ALL)		
MEDIAN(FSNUFFRR,ALL)	.301	.605
MEDIAN(FAPNOERR,ALL)		-.348

Extraction Method: Principal Component Analysis.

a. 2 components extracted.

Rotated Component Matrix[a]

	Component	
	1	2
MEDIAN(SACTIVER,ALL)	.783	
MEDIAN(SSTILLR,ALL)	.762	
MEDIAN(SSTRIDRR,ALL)	.738	.416
MEDIAN(FSTILLRR,ALL)	.725	
MEDIAN(FACTIVRR,ALL)	.706	
MEDIAN(SWHEEZER,ALL)	.703	.480
MEDIAN(SFASTERR,ALL)	.653	.505
MEDIAN(FWHEEZRR,ALL)	.597	.437
MEDIAN(FSTRIRR,ALL)	.587	.408
MEDIAN(FFASTRR,ALL)	.580	.502
MEDIAN(SSLOWERR,ALL)	.573	
MEDIAN(FSLOWRR,ALL)	.528	
MEDIAN(FAPNOERR,ALL)	.331	
MEDIAN (FNTCOUGR, ALL)		.775
MEDIAN(SNTCOUGR,ALL)	.382	.771
MEDIAN(SDTCOUGR,ALL)	.415	.762
MEDIAN(FDTCOURR,ALL)		.736
MEDIAN(SSNUFFLR,ALL)		.687
MEDIAN(FSNUFFRR,ALL)		.657
MEDIAN(SAPNOEAR,ALL)		

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 3 iterations.

Component Transformation Matrix

Component	1	2
1	.769	.639
2	-.639	.769

Extraction Method: Principal Component Analysis.
Rotation Method: Varimax with Kaiser Normalization.

Factor 1 = 'miscellaneous symptom' factor
Factor 2 = 'cough and snuffles' factor

Appendix XIV – PAF of IPQ items from merged Phase II/III data

PAF (1-factor solution) 16 IPQ items merged Phase II/III data – SPSS Output

Communalities

	Initial	Extraction
Illness would last a short time - recoded	.460	.244
Illness would be permanent rather than temporary	.440	.276
Illness would last a long time	.541	.484
Illness was serious	.265	6.996E-02
Illness has had consequences on child's life	.471	.246
Illness has become easier to live with - recoded	.254	4.140E-03
Illness has had littel effect on child's life - recoded	.460	.190
Illness has affected the way others see child	.319	.190
Illness has had economic consequences	.270	.254
Illness has affected the way I see my child	.272	5.121E-02
Illness will improve with time - recoded	.334	1.849E-02
Child could control symptoms	.226	3.263E-03
Little could be done to improve illness - recoded	.320	2.640E-02
Treatment would be effective	.277	2.571E-03
Child's recovery due to chance - recoded	.290	.119
Child could control his illness	.240	8.860E-02

Extraction Method: Principal Axis Factoring.

Total Variance Explained

	Initial Eigenvalues			Extraction Sums of Squared Loadings		
Factor	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	3.013	18.834	18.834	2.268	14.176	14.176
2	1.889	11.805	30.638			
3	1.672	10.452	41.090			
4	1.361	8.503	49.593			
5	1.274	7.961	57.554			
6	1.220	7.625	65.178			
7	1.020	6.375	71.554			
8	.795	4.971	76.525			
9	.699	4.368	80.892			
10	.604	3.772	84.664			
11	.573	3.582	88.247			
12	.497	3.106	91.353			
13	.430	2.685	94.038			
14	.385	2.408	96.446			
15	.289	1.805	98.252			
16	.280	1.748	100.000			

Extraction Method: Principal Axis Factoring.

Factor Matrix[a]

	Factor
	1
Illness would last a long time	.696
Illness would be permanent rather than temporary	.526
Illness has had economic consequences	.504
Illness has had consequences on child's life	.496
Illness would last a short time	.494
Illness has had little effect on child's life	.436
Illness has affected the way others see child	.436
Child's recovery due to chance	-.345
Child could control his illness	
Illness was serious	
Illness has affected the way I see my child	
Little could be done to improve illness	
Illness will improve with time	
Illness has become easier to live with	
Child could control symptoms	
Treatment would be effective	

Extraction Method: Principal Axis Factoring.

a. 1 factors extracted. 5 iterations required.

PAF (2-factor solution) 16 IPQ items merged Phase II/III data (varimax rotation) – SPSS output

Communalities

	Initial	Extraction
Illness would last a short time - recoded	.460	.426
Illness would be permanent rather than temporary	.440	.393
Illness would last a long time	.541	.655
Illness was serious	.265	8.874E-02
Illness has had consequences on child's life	.471	.305
Illness has become easier to live with - recoded	.254	2.390E-02
Illness has had littel effect on child's life - recoded	.460	.179
Illness has affected the way others see child	.319	.313
Illness has had economic consequences	.270	.284
Illness has affected the way I see my child	.272	.293
Illness will improve with time - recoded	.334	.159
Child could control symptoms	.226	3.905E-02
Little could be done to improve illness - recoded	.320	3.293E-02
Treatment would be effective	.277	3.308E-02
Child's recovery due to chance - recoded	.290	.116
Child could control his illness	.240	.204

Extraction Method: Principal Axis Factoring.

Note: E-02 = 10^{-2}

Total Variance Explained

	Initial Eigenvalues			Extraction Sums of Squared Loadings			Rotation Sums of Squared Loadings		
Factor	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	3.013	18.834	18.834	2.372	14.824	14.824	2.046	12.787	12.787
2	1.889	11.805	30.638	1.174	7.339	22.163	1.500	9.375	22.163
3	1.672	10.452	41.090						
4	1.361	8.503	49.593						
5	1.274	7.961	57.554						
6	1.220	7.625	65.178						
7	1.020	6.375	71.554						
8	.795	4.971	76.525						
9	.699	4.368	80.892						
10	.604	3.772	84.664						
11	.573	3.582	88.247						
12	.497	3.106	91.353						
13	.430	2.685	94.038						
14	.385	2.408	96.446						
15	.289	1.805	98.252						
16	.280	1.748	100.000						

Extraction Method: Principal Axis Factoring.

Factor Matrix[a]

	Factor	
	1	2
Illness would last a long time	.739	-.331
Illness would be permanent rather than temporary	.546	-.308
Illness would last a short time	.528	-.385
Illness has had economic consequences	.498	
Illness has had consequences on child's life	.489	
Illness has affected the way others see child	.447	.337
Illness has had littel effect on child's life	.421	
Child's recovery due to chance	-.336	
Illness was serious		
Little could be done to improve illness		
Illness has affected the way I see my child		.485
Illness will improve with time		-.371
Child could control his illness	.302	.336
Child could control symptoms		
Treatment would be effective		
Illness has become easier to live with		

Extraction Method: Principal Axis Factoring.

a. 2 factors extracted. 9 iterations required.

Rotated Factor Matrix[a]

	Factor	
	1	2
Illness has affected the way others see child	.557	
Illness has had consequences on child's life	.551	
Illness has had economic consequences	.524	
Illness has affected the way I see my child	.459	
Child could control his illness	.433	
Illness has had little effect on child's life	.380	
Child's recovery due to chance	-.317	
Illness was serious		
Little could be done to improve illness		
Child could control symptoms		
Illness would last a long time	.458	.668
Illness would last a short time		.604
Illness would be permanent rather than temporary	.305	.548
Illness will improve with time		.393
Treatment would be effective		
Illness has become easier to live with		

Extraction Method: Principal Axis Factoring.
Rotation Method: Varimax with Kaiser Normalization.

a. Rotation converged in 3 iterations.

Note: Factor loading scores less than 0.3 are shown as blank in the above table

Factor Transformation Matrix

Factor	1	2
1	.853	.522
2	.522	-.853

Extraction Method: Principal Axis Factoring.
Rotation Method: Varimax with Kaiser Normalization.

Definitions

Abbreviations

ADL	Activities of Daily Living
AIC	Adult Intensive Care
AICU	Adult Intensive Care Unit
ALL	Acute Lymphoblastic Leukaemia
AMA	About My Asthma
ANOVA	Analysis of Variance
AUQEI	Autoquestionnaire Enfant Imagé
BIDS	Bath Information and Data Services
CAQ	Childhood Asthma Questionnaire
CAQA	Childhood Asthma Questionnaire Form A
CAQB	Childhood Asthma Questionnaire Form B
CAQC	Childhood Asthma Questionnaire Form C
CCTR	Cochrane Controlled Trials Register
CDSR	Cochrane Database of Systematic Reviews
CF	Cystic Fibrosis
CGAS	Children's Global Assessment Scale
CGI	Clinician-generated Index
CHQ	Child Health Questionnaire
CHSI	Child Health Status Index
CINAHL	Cumulative Index of Nursing and Allied Health Literature
ComQol	Comprehensive quality of life
COOP	Dartmouth Primary Care Cooperative
C-QOL	Child Quality of Life
CQOL	Child Quality of Life
CRD	Centre for Reviews and Dissemination
CRMD	Cochrane Review Methodology Database
DAI	Dissertation Abstracts International
DARE	Database of Abstracts of Reviews of Effectiveness
DGH	District General Hospital
DIF	Differential Item Functioning
ECMO	Extracorporeal Membrane Oxygenation
EHRQL	Exeter Health-related Quality of Life

Abbreviations (continued)

ELBW	Extremely Low Birth Weight
EMBASE	Excerpta Medica Online
FA	Factor Analysis
FDI	Functional Disability Inventory
FS	Functional Status
GHRI	General Health Rating Index
GQL	Generic Child Quality of Life
GOS	Great Ormond Street
GP	General Practitioner
HAD	Hospital Anxiety Depression
HAY	How Are You
HELMIS	Health Management Information Service
HIE	Health Insurance Study
HIS	Health Insurance Experiment
HIV	Human Immunodeficiency Virus
HMIC	Health Management Information Consortium
HRQoL	Health-related Quality of Life
HTA	Health Technology Assessment
HUI	Health Utilities Index
HV	Health Visitor
ICU	Intensive Care Unit
ICC	Intraclass Correlation
ICNARC	Intensive Care National Audit Research Centre
INAHTA	International Network of Agencies for Health Technology Assessment
IPQ	Illness Perception Questionnaire
ISI	Institute for Scientific Information
ITC	Item Total Correlation
ITQoL	Infant Toddler Quality of Life
KINDL	No definition
LBW	Low-Birth Weight
LREC	Local Research Ethics Committee
MEDLINE	Index Medicus Online
MM	Myelomeningocele

Abbreviations (continued)

MMQL	Minneapolis-Manchester Quality of Life
NHIS	National Health Interview Survey
NHS	National Health Service
NHS CRD	NHS Centre for Reviews and Dissemination
NHSE	NHS Executive
NICU	Neonatal Intensive Care Unit
NIC	Neonatal Intensive Care
NRR	National Research Register
NUD*IST	Non-numerical Unstructured Data Indexing
OPCS	Office of Population Censuses and Surveys
OSA	Obstructive Sleep Apnoea
PAF	Principal Axial Factoring
PAQLQ	Pediatric Asthma Quality of Life Questionnaire
PedsQL	Pediatric Quality of Life
PCA	Principal Components Analysis
PCCP	Paediatric Cerebral Performance Category
PCQL	Pediatric Cancer Quality of Life
PEDI	Paediatric Evaluation of Disability Inventory
PIC	Paediatric Intensive Care
PICQoL	Paediatric Intensive Care Quality of Life
PICS	Paediatric Intensive Care Society
PICU	Paediatric Intensive Care Unit
PIM	Paediatric Index of Mortality
POPC	Paediatric Overall Performance Category
PRISM	Paediatric Risk of Mortality
PsychLit	Psychological Literature Database
PsycINFO	Psychological Information Database
PTSD	Post Traumatic Stress Disorder
QLQC	Quality of Life Questionnaire for Children
QoL	Quality of life
QOLPAV	Quality of Life Profile – Adolescent Version
QSR	Qualitative Solutions and Research
QUALIN	Infant Quality of Life Questionnaire

Abbreviations (continued)

QWB	Quality of Wellbeing
RAG	Research Advisory Group
RAHC MOF	Royal Alexander Hospital for Children Measure of Function
RAND	No definition
RCT	Randomised controlled trial
SEIQoL	Schedule for the Evaluation of Individual Quality of Life
SF-36	Short-form 36
SIGLE	System for Information on Grey Literature
SMR	Standardised Mortality Ratio
SRM	Standardised Response Mean
STAI	State Trait Anxiety Inventory
TACQOL	No definition
TAPQOL	TNO-AZL Preschool Children Quality of Life
TISS	Therapeutic Intervention Scoring System
UK	United Kingdom
USA	United States of America
VSP-A	Vécu et Santé Perçue de l'Adolescent
WCHMP	Warwick Child Health and Morbidity Profile
Wee-FIM	Functional Independence Measure for Children
WHO	World Health Organisation
WHOQOL	World Health Organisation Quality of Life Group

Glossary

This glossary has been adapted from one described by Jenkinson and McGee (1998) and utilises definitions described within the thesis.

Acquiescence A form of satisficing where a respondent tends to give positive responses, and agree with every statement, also known as yes-saying.

Appropriateness Refers to whether a measure is appropriate to the target population and setting.

Ceiling and floor effects Refer to the response range and the method of scoring a measure. A measure applied to a random sample of the population which is not sensitive to the lower levels of ill health and that is scored from 0 (good health) to 100 (poor health) would be said to manifest a floor effect, as most respondents would score 0. If the measure were scored from 0 (poor health) to 100 (good health) this would be referred to as a ceiling effect, as most respondents would score 100. Floor and ceiling effects are likely to be found in measures with small numbers of items.

Clinical effectiveness The extent to which specific clinical interventions, when deployed in the field for a particular patient or population, do what they are intended to do, that is, maintain and improve health and secure the greatest possible health gain from the available resources (including available evidence).

Clinical governance A framework through which NHS organisations are accountable for continuously improving the quality of their services and safeguarding high standards of care by creating an environment in which excellence in clinical care will flourish.

Clinical trial An experiment to assess the efficacy of a treatment.

Clinical utility The ability of a measure to be used in routine clinical practice.

Construct A phenomenon that exists theoretically but that cannot be measured directly and is defined or operationalised in terms of other observable indicators. It can be thought of as a 'mini theory' to explain the relationships among various behaviours or attitudes.

Glossary (continued)

Construct validity Linking the attribute that is being measured to some other attribute by a hypothesis or construct. The hypotheses will explore the difference between two or more populations who would be expected to have differing amounts of the property assessed by the measure. The hypothetical construct is tested by applying the measure to the appropriate samples; if the expected relationship is found, then the hypothesis and the measure are sound, if no relationship is found, the fault may lie with either the measure or the hypothesis.

Content validity The extent to which items on a questionnaire tap all relevant aspects of the attribute they are intending to measure.

Convergent and discriminant validity A measure should both converge with other indicators of the same concept and be able to discriminate unrelated indicators.

Cost-effectiveness analysis A form of economic evaluation where the consequences of programmes are measured in the most appropriate natural effects or physical units, such as 'years of life gained' or 'cases correctly diagnosed'; no attempt is made to value the consequences, so it is implicitly assumed that the output concerned is in some sense 'worth having'.

Cost-utility analysis A form of economic cost-effectiveness analysis where the effects of health care interventions are assessed according to the quality-adjusted life-years gained or lost.

Criterion validity The extent to which a measure correlates with a pre-existing one, preferable a 'gold standard'. There are two types: 1) concurrent validity, where a new measure is administered at the same time as a pre-existing one; and 2) predictive validity.

Cronbach's alpha statistic A statistic used to determine the internal reliability of a measure based on the mean correlation between items.

Deduction Process of data gathering to test predefined theory or hypotheses.

Glossary (continued)

Deviation Tendency to respond to items in a questionnaire with deviant responses.

Dimensions (domains) of health Theoretically or empirically distinct aspects of health, e.g. physical, mental or social health.

Disease-specific measures Questionnaires designed for use with a particular patient group, e.g. the Childhood Asthma Questionnaire was designed for specific use with children with asthma.

Economic evaluation The comparative analysis of alternate courses of action in terms of both their costs and consequences.

Effect size A statistic for determining the difference between scores gained at two different times by dividing the mean change in score by the baseline standard deviation.

End aversion bias Also known as central tendency bias, where some respondents are reluctant to use extreme categories of a scale.

Evidence based practice The conscientious, explicit, and judicious use of current best evidence, based on systematic review of all available evidence – including patient-reported, clinician-observed and research-derived evidence – in making and carrying out decisions about the care of individual patients.

Face validity The need for a questionnaire to apparently tap, simply by item content, an underlying dimension; questions should be unambiguous and easily understood and should reflect issues appropriate to the dimension.

Faking bad When a respondent is aware and is intentionally attempting to create a false negative impression on a questionnaire.

Faking good When a respondent is aware and is intentionally attempting to create a false positive impression on a questionnaire.

Glossary (continued)

Factor analysis A group of statistical techniques whose purpose is to reduce a large number of variables to a smaller number of latent variables – variables that can be measured directly.

Feasibility The ability to use the measure within the application area

Framing When a respondent's choice between two alternative states depends on how these states are framed or worded.

Functional status An individual's ability to perform normal activities to meet basic needs, fulfil usual roles and maintain health and wellbeing.

Generic measure A measure designed for use with any illness groups or populations samples, as opposed to those intended for specific illness groups.

Halo effect Where instead of assessing each item separately, respondents let themselves be influenced by their general feelings of like or dislike.

Health Defined by the World Health Organisation is a state of complete physical, mental, and social wellbeing and not merely the absence of disease or infirmity.

Health index Where all the items in an instrument are summed producing one overall score.

Health outcomes The end results of clinical interventions and processes; these can be assessed in terms of mortality, morbidity, physiological measures, and subjective patient-based assessments of health.

Health profile A questionnaire covering various dimensions of health, as opposed to a health index which sums all measured aspects of health into a single figure.

Glossary (continued)

Health-related quality of life (HRQoL) An individual's level of health-related wellbeing; measurement of HRQoL addresses various dimensions of health and is often multidimensional.

Health services research A scientific activity to test hypotheses and thus generate new knowledge that may subsequently be useful in improving the effectiveness or efficiency of health care.

Health status A level of health in terms of physical, social and mental wellbeing measured at a particular point in time.

Index of responsiveness A statistic for determining the difference between scores gained at two different times.

Induction Process of moving from data towards generalisations, hypotheses or theory.

Internal consistency/reliability Involves examining the extent to which a number of items addressing the same concept actually are doing so. The correlation can be calculated between items using split-half reliability, whereby the measure is randomly split into two groups and reliability is assessed by the correlation between the two half tests, or by Cronbach's alpha statistic (see Cronbach's alpha statistic).

Interpretability Asks how meaningful scores of HRQoL are to clinicians and patients.

Inter-rater reliability The consistency of a measure when administered by different interviewers within a short period of time. The Kappa statistic calculates a coefficient of agreement to assess whether differences were due to agreement or chance.

Item An individual question which may stand alone or form part of a battery of questions in a dimension.

Item content The actual wording of the individual questions, which must satisfy the requirements of face validity.

Glossary (continued)

Latent variable A variable that cannot be measured directly, but is measured indirectly.

Likert scale A response scale in which respondents select from a range of options which are placed on a continuum, such as 'Excellent', Very Good', 'Good', 'Fair', and 'Poor.'

Longitudinal study Where individuals in a study are followed over time.

Multidimensional measures Measures which consider health in more than one dimension or domain.

Non-parametric methods Statistical analyses that assume that data do not follow the normal distribution.

Normal distribution Data that produce a 'bell curve' with the mean, mode, and median all having the same value.

Normative data Data that are representative of a population

Optimising Performance of tasks when completing an item on a questionnaire to include interpreting the meaning of the question, retrieving all relevant information from memories, forming a single, integrated summary judgement, and conveying that judgement on the answer sheet.

Paediatric intensive care Delivers care to the most critically ill children, most of who will be artificially ventilated. It is a low volume, high cost service, cannot be provided in every locality, requires highly trained specialist staff, and should be available to all children who need it regardless of where they live.

Paediatric intensive care unit A PICU is a centre where children who become critically ill at any time between early infancy and adolescence (usually up to 16 years of age) are treated.

Glossary (continued)

Paradigm A pattern that may serve as a model or example; paradigms encompass both theories and methods, e.g. quantitative or qualitative paradigm.

Parametric methods Statistical analyses in which data are assumed to be normally distributed.

Patient outcome A final health status measurement after a passage of time and the application of a treatment.

Patient-based outcome Outcomes identified and valued by patients.

Precision The ability of a measure to differentiate between illness groups or states of health.

Positive skew Where responses on a questionnaire are not evenly distributed over the range of alternatives, but show a positive skew toward the favourable end.

Predictive validity The ability of a measure to predict some other measure of outcome.

Psychometrics Sciences of measuring mental and subjective phenomena.

Purposive sample Deliberate choice of respondents or settings.

Quality of life (QoL) A multidimensional construct encompassing several dimensions, including physical status and functional abilities, psychological status and wellbeing, social interactions, economic and/or vocational status and factors, and religious and/or spiritual status. It differs from the term health-related quality of life which is the impact of disease, illness or injury, medical treatment or health care policy on an individual; often it excludes income, freedom, and quality of the environment, that do not directly affect health issues.

Glossary (continued)

Quality-adjusted-life-year (QALY) A generic measure of health benefit which attempts to represent the relative value attached by society to different improvements in health, enabling systematic comparison between a variety of health care interventions. Comparisons between treatment programmes are expressed in QALYs. With a measure of both the life-years gained from a particular intervention and the quality of life in each of those years it is possible to calculate the number of QALYs obtained. An index of quality of life multiplied by the number of years in each health state equals the number of QALYs.

Random sample Each individual in the given population has an equal chance of selection into the sample.

Reliability A reliable measure is one that produces consistent results from the same participants at different times when no evidence of change exists.

Response range The set of answers available to respondents for each item in a questionnaire.

Responsiveness The extent to which a measure can detect change in health status over time.

Satisficing Giving an answer on a questionnaire which is satisfactory, but not optimal.

Sensitivity to change A measure's ability to detect change over time, also known as responsiveness.

SF-36 Short-form 36 is a generic adult health profile developed from RAND Corporation's Health Insurance experiment and subsequent Medical Outcomes Study. A shorter version is available, the SF-12.

Social desirability Respondents tend to give a socially desirable answer on a questionnaire; the respondent is not deliberately trying to deceive or lie.

Glossary (continued)

Standardised mortality ratio The ratio of observed mortality and expected mortality adjusted for severity of illness.

Standardised response mean A statistic for determining the difference between scores gained at two different times. It is calculated by dividing the mean change on a scale by the mean change in the standard deviation.

Subjective wellbeing The patient's assessment of his or her own health status as opposed to professionally or clinically defined indicators. For children, this assessment may be via a proxy.

Systematic review A review in which methods for selecting and including or excluding publications are explicitly stated.

Tertiary health care A specialised, highly technical level of health care including the diagnosis and treatment of disease and disability in sophisticated, large research and teaching hospitals.

Test-retest reliability The administration of a measure on two separate occasions to the same population; the correlation between scores provides an estimate of the reliability of the measure. The two occasions need to be far enough apart so that the previous responses cannot be remembered but close enough in time so that change in the true score is minimal.

Thurstone's method of equal appearing intervals A comparative method of scaling where a set of items is calibrated so that they can be placed on an interval scale. Responses to these items are then used in developing a score by summing or averaging the calibration weights of those items endorsed by a respondent. Thurstone scaling begins with a large number of items, and respondents are asked to judge each item against all others by explicitly ranking the items.

User-centredness The extent to which a measure faithfully captures both the content of the patient's views and the form in which their views are expressed.

Glossary (continued)

Utility The preference for or desirability of a particular outcome in terms of health status.

Validity The extent to which a measure measures the desired underlying concept.

Visual analogue scale Typically a 10cm line on which the respondent indicates the intensity of his or her response; phrases are printed at the ends of the line, such as 'no pain' or 'extreme pain' to indicate the scope of the scale.

Weighting Items, which are given values indicating their relative importance to other items on a scale, are said to be weighted.

References

Acquadro C, Jambon B, Ellis D & Marquis P (1996) Language and translation issues. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 575-585.

Adams PF, Hardy AM (1989) Current estimates from the National Health Interview Survey: United States, 1988. National Center for Health Statistics. *Vital Health Statistics,* 10 (173).

Anderson RT, McFarlane M, Naughton MJ & Shumaker SA (1996) Conceptual issues and considerations in cross-cultural validation of generic health-related quality of life instruments. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 605-612.

Apajasalo M, Rautonen J, Holmberg C & Sinkkonen J (1996a) Quality of life in pre-adolescence: a 17-dimensional health-related measure (17D). *Quality of Life Research,* 5 (6): 532-538.

Apajasalo M, Sinotonen H, Holmberg C, Sinkkonen J, Aalberg V, Pihko H, Siimes A, Kaitila I, Makela A, Rantakari K, Anttila R & Rautonen J (1996b) Quality of life in early adolescence: A sixteen-dimensional health-related measure (16D). *Quality of Life Research,* 5, 205-211.

Apajasalo M, Rautonen J, Sintonen H & Holmberg C (1997) Health-related quality of life after organ transplantation in childhood. *Pediatric Transplantation,* 1 92): 130-137.

Apajasalo M, Sintonen H, Rautonen J & Kaitila I (1998) Health-related quality of life of patients with genetic skeletal dysplasias. *European Journal of Pediatrics,* 157 (2): 114-121.

Asmussen L, Olson LM, Grant EN, Landgraf JM, Fagan J & Weiss, KB (2000) Use of the child health questionnaire in a sample of moderate and low-income inner-city children with asthma. *American Journal of Respiratory Critical Care Medicine,* 162 94): 1215-1221.

Baker M (1998) *Making Sense of the new NHS White Paper,* Radcliffe Medical Press: Oxon.

Barr RD, Pai MKR, Weitzman S, Feeny D, Furlong W, Rosenbaum P & Torrance GW (1994). A multi-attribute approach to health status measurement and clinical management – illustrated by application to brain tumors in childhood. *International Journal of Oncology,* 4: 639-648.

Barr RD, Petrie C, Furlong WJ, Rothney M & Feeny D (1997) Health-related quality of life during post-induction chemotherapy in children with acute lymphoblastic leukaemia in remission. *International Journal of Oncology,* 11: 333-339.

Barr RD, Simpson T, Whitton A, Rush B, Furlong W & Feeny DH (1999) Health-related quality of life in survivors of tumours of the central nervous system in childhood – a preference-based approach to measurement in a cross-sectional study. *European Journal of Cancer,* 35 (2): 248-255.

Barry P & Hocking M (1995) Paediatric intensive care in the UK: who gets admitted and why? *British Journal of Intensive Care,* 5 (7): 227-232.

Bekker HL, Legare F, Stacey D, O'Connor A & Lemyre L (2003) *(In Press)* Is anxiety an appropriate measure of decision aid effectiveness: a systematic review? *Patient Education and Counselling.*

Bergner MB (1985) Measurement of health status. *Medical Care,* 23 (5): 696-704.

Billson AL & Walker DA (1994) Assessment of health status in survivors of cancer. *Archives of Disease in Childhood,* 70: 200-204.

Black N (1996) Why we need observational studies to evaluate the effectiveness of health care. *BMJ,* 312: 1215-1218.

Blacktop J (1996) A discussion of different sampling techniques. *Nurse Researcher,* 3 (4): 5-15.

Bland M (1997) *An Introduction to Medical Statistics* (2nd Edition), Oxford University Press: Oxford.

Bonn M (1994) The effects of hospitalisation on children: a review. *South African Journal of Nursing,* 17: 20-24.

Bouman NH, Koot HM, Van Gils APJM & Verhulst F (1999) Development of a health-related quality of life instrument for children: the Quality of Life Questionnaire for Children. *Psychology and Health,* 14: 829-846.

Bowling A (1995) *Measuring Disease,* Open University Press: Buckingham.

Bowling A (1997a) *Measuring Health. A Review of Quality of Life Measurement Scales* (2nd Edition), Open University Press: Buckingham.

Bowling A (1997b) R*esearch Methods in Health – Investigating Health and Health Services,* Open University Press: Buckingham.

Boyle MH, Torrance GW, Sinclair JC et al (1983) Economic evaluation of neonatal intensive care of very-low-birth-weight infants. *New England Journal of Medicine,* 308 (22): 1330-1337.

Boyle MH, Furlong W, Feeny D, Torrance GW & Hatcher J (1995) Reliability of the Health Utilities Index – Mark III used in the 1991 cycle 6 Canadian General Social Survey Health Questionnaire. *Quality of Life Research,* 4 (3): 249-257.

BPA (1993) *The Care of Critically Ill Children – Report of a Multidisciplinary Working Party on Intensive Care.* BPA: London.

Bradlyn AS, Harris CV, Warner JE, Ritchey AK and Zaboy K (1993) An investigation of the validity of the Quality of Well-Being Scale with pediatric oncology patients. *Health Psychology,* 12(3): 246-250.

Bray RJ & Morrell P (1982) A follow-up of the survivors of mechanical ventilation in a paediatric intensive care unit. *Intensive Care Medicine,* 8: 163.

Brazier J & Deverill M (1998) The use of health-related quality of life measures in economic evaluation, In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods. A Guide to Best Practice,* BMJ Books: London, 23-34.

Brettle AJ, Long AF, Grant MJ & Greenhalgh J (1998) Searching for information on outcomes: do you need to be comprehensive? *Quality in Health Care,* 7: 163-167.

Bridge PD, Lee H & Silverman M (1996) A portable device based on the interrupter technique to measure bronchodilator response in schoolchildren. *European Respiratory Journal,* 9 (7): 1368-1373.

Brink PJ (1991) Issues of reliability and validity. In: Morse J (Ed) *Qualitative Nursing Research,* Sage Publications Inc: London, 164-187.

Britten N (1996) Qualitative interviews in medical research. In: Mays N & Pope C (Eds) *Qualitative Research in Health Care,* BMJ Publishing Group: London, 28-35.

Bruill J, Maes S, Le Coq L & Boeke J (1997) The Development of the How Are You? (HAY) a quality of life questionnaire for children with a chronic illness. *Quality of Life Newsletter,* 18.

Bryman A & Cramer D (2001) *Quantitative Data Analysis with SPSS 10 for Windows – A Guide for Social Scientists,* Routledge: Hove.

Bukstein DA, McGrath MM, Buchner DA, Landgraf J & Goss TF (2000) Evaluation of a short form for measuring health-related quality of life among asthma patients. *Journal of Allergy and Clinical Immunology,* 105 (2): 245-251.

Bullinger M & Ravens-Sieberer U (1995) Health related quality of life assessment in children: a review of the literature. *Revue Européenne de Psychologie Appliquée,* 45 (4): 245-254.

Butt W, Shann F, Tibballs J, Williams J, Cuddihy L, Blewett L, & Farley M (1990) Long-term outcome of children after intensive care. *Critical Care Medicine,* 18 (9): 961-965.

Cadman D, Goldsmith C & Bashim P (1984) Values, preferences and decisions in the care of children with developmental disabilities. *Journal of Developmental and Behavioural Pediatrics,* 5: 60-64.

Cadman D, Goldsmith C, Torrance G, Boyle M & Furlong W (1986) *Development of a Health Status Index for Ontario Children, Final Report to the Ontario Ministry of Health on Research Grant DM648 (006633),* McMaster University: Hamilton.

Carnevale FA (1988) Behavioural rating of anxiety in parents of critically ill children. *Journal of Palliative Care,* 4 (3): 72.

Carnevale FA (1990) A description of stressors and coping strategies among parents of critically ill children – a preliminary study. *Intensive Care Nursing,* 6: 4-11.

Catell RB (1952) *Factor Analysis: An Introduction and Manual for the Psychologist and Social Scientist,* Harper: New York.

Cavanagh S (1997) Content analysis: concepts, methods and applications. *Nurse Researcher,* 4 (3): 16.

CDSR (2000) [database available on-line]. NHS Centre for Reviews and Dissemination, University of York: York.

Chalmers TC, Franck CS & Reitman D (1990) Minimising the three stages of publication bias. *JAMA,* 263 (10): 1392-1395.

Cheater F (1998) Quality of life measures for the healthcare environment. *Nurse Researcher,* 5 (3): 17-30.

Chowienczyk P J, Lawson C P, Lane S, Johnson R, Wilson N, Silverman M & Cochrane G M (1991) a flow interruption device for the measurement of airway resistance. *European Respiratory Journal,* 4 (5): 623-628.

Christie MJ, French DJ, Sowden A & West A (1993) The development of child-centred, disease-specific, questionnaires for contemporary appraisals of living with asthma. *Psychosomatic Medicine,* 55: 541-548.

Clarke E, Sulaiman S, Tim CF, Chi SLP, Mital R & Lee BW (1999) Pediatric Asthma quality of Life Questionnaire: Validation in children from Singapore. *Asian Pacific Journal of Allergy & Immunology,* 17 (3): 155-161.

Clegg F (1990) *Simple Statistics – A Course Book for Social Sciences,* Cambridge University Press: Cambridge.

Cobb A & Hagemaster J (1987) Ten criteria for evaluating qualitative research proposals. *Journal of Nursing Education,* 26: 138-143.

Cohen J (1960) A coefficient of agreement for nominal scales. *Educational and Psychological Measurement,* 20: 37-46.

Colclough J (1998) *HMIC via ARC,* University of York: York.

Colclough J (1999) *Supplementary Sources in Health Sciences,* University of York: York.

Colclough J (2000a) *CINAHL via ARC,* University of York: York.

Colclough J (2000b) *EMBASE via ARC,* University of York: York.

Collier J & MacKinlay D (1997) Developing a generic child quality of life measure, H*ealth Psychology Update,* 28: 12-16.

Collier J, MacKinlay D & Phillips D (2000) Norm values for the generic children's quality of life measure (GCQ) from a large school-based sample. *Quality of Life Research,* 9 (6): 617-623.

Cook DJ, Guyatt GH, Ryan G, Clifton J, Buckingham L, Willan A, McIlroy W & Oxman AD (1993) Should unpublished data be included in meta-analyses? Current convictions and controversies. *Journal of the American Medical Association,* 269: 2749-2753.

Cooper C (2000) The use of electronic mail in the research process. *Nurse Researcher,* 7 (4): 24-30.

Cormack D (1991) *The Research Process in Nursing,* Oxford: Blackwell Scientific Publications.

Cox P (1992) Children in critical care: how parents cope. *British Journal of Nursing,* 1 (15): 764-768.

Creswell JW (1994) *Research Design – Qualitative and Quantitative Approaches,* Thousand Oaks, California: Sage Publications.

Cronbach LJ (1951) Coefficient alpha and the internal structure of tests. *Psychometrika,* 16: 297-334.

Cronbach LJ & Meehl PE (1955) Construct validity in psychological tests. *Psychological Bulletin,* 52: 281-302.

Cullen DJ, Civetta JM & Briggs BA et al, (1974) Therapeutic intervention scoring system: A method for quantitative comparison of patient care. *Critical Care Medicine,* 2: 57.

Cullum N (1998) Clinical effectiveness in nursing. *Nursing Times Research,* 3 (1): 15-18.

Curley MAQ (1996) The essence of pediatric critical care nursing In: Curley MAQ (Ed) *Critical Care Nursing of Infants and Children,* WB Saunders Company: Philadelphia, 3-14.

DARE (2000) [database available on-line]. University of York: NHS Centre for Reviews and Dissemination.

Davenport GC (1994) *An Introduction to Child Development* (2nd Edition), CollinsEducational: London.

De Keizer NF, Bonsel GJ & Gemke RJBJ (1997) Health status prediction in critically ill children: A pilot study introducing Standardized Health Ratios. *Quality of Life Research,* 6: 192-199.

De Keizer NF, Bonsel GJ & Gemke RJBJ (1998) TISS and real paediatric ICU costs: a case study with generalisable methodology. *Intensive Care Medicine,* 24 (10): 1062-1069.

Dey I (1993) *Qualitative Data Analysis – A User Friendly Guide for Social Scientists,* Routledge: London.

Dickersin K, Scherer R & Lefebvre C (1994) Systematic reviews: identifying relevant studies for systematic reviews. *BMJ,* 309: 1286-1291.

DoH (1998) *A First Class Service – Quality in the new NHS,* Department of Health: London.

Donovan JL, Frankel SJ & Eyles JD (1993) Assessing the need for health status measures. *Journal of Epidemiology and Community Health,* 47: 158-162.

Dosseter DR, Liddle JLM & Mellis CM (1996) Measuring health outcome in paediatrics: Development of the RAHC measure of function. *Journal of Pediatrics in Child Health,* 32: 519-524.

Downes JJ (1992) The historical evolution, current status, and prospective development of pediatric critical care. *Critical Care Clinics of North America,* 8 (1): 1-22.

Drotar D, Levi R, Palermo TM, Riekert KA, Robinson JR & Walders N (1998a) Recommendations for research concerning the measurement of pediatric health-related quality of life. In: Drotar D (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents – Implications for Research and Practice,* Mahwah: Lawrence Erlbaum Associates, 341-349.

Drotar D, Levi R, Palermo TM, Riekert KA, Robinson JR & Walders N (1998b) Clinical applications of health-related quality of life assessment for children and adolescents. In: Drotar D (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents – Implications for Research and Practice,* Mahwah: Lawrence Erlbaum Associates, 329-339.

Drummond MF, O'Brien B, Stoddart GL & Torrance GW (1997) *Methods of Economic Evaluation of Health Care Programmes,* Oxford Medical Publications: Oxford.

Eberly TW, Miles MS, Carter MC, Hennessey J & Riddle II (1985) Parental stress after the unexpected admission of a child to the intensive care unit. *Critical Care Quarterly,* 8 (1): 57-65.

Ebbs SR, Fallowfield LJ, Fraser SCA & Baum M (1989) Treatment outcomes and quality of life. *International Journal of Technology Assessment in Health Care,* 5: 391-400.

Edbrooke DL, Wilson AJ, Gerrish P & Mann AJ (1995) The Sheffield costing system for intensive care, *Care of the Critically Ill,* 11 (3): 106-110.

Edwards SJL, Lilford RJ & Kiauka S (1998) Different types of systematic review in health services research. In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods – A Guide to Best Practice,* BMJ Books: London, 255-259.

Edwards A & Talbot R (1994) *The Hard-pressed Researcher – A Research Handbook for the Caring Professions,* Longman: London.

Eisen W, Ware JE, Donald CA & Brook RH (1979) Measuring components of children's health status. *Medical Care*, 17(9): 902-921.

Eiser C (1995) Choices in measuring quality of life in children with cancer: A comment. *Psychooncology,* 4: 121-131.

Eiser C (1997) Children's quality of life measures. *Archives of Diseases in Childhood,* 77: 350-354.

Eiser C, Cotter I, Oades P, Seamark D & Smith R (1999) Health-related quality of life measures for children. *International Journal of Cancer,* S12: 87-90.

Eiser C, Havermans T, Craft A & Kernahan J (1995) Development of a measure to assess the perceived illness experience after treatment for childhood cancer. *Archives of Disease in Childhood,* 72: 302-307.

Eiser C, Kopel S, Cool P & Grimer R (1999) The Perceived Illness Experience scale (PIE): reliability and validity revisited. *Child Care Health Development,* 25 (3): 179-190.

Eiser C, Mohay H & Morse R (2000) The measurement of quality of life in young children. *Child: Care, Health and Development,* 26 (5): 401-414.

Eiser C & Morse R (2001a) Quality-of-life measures in chronic diseases of childhood. *Health Technology Assessment,* 5 (4).

Eiser C & Morse R (2001b) Can parents rate their child's health-related quality of life? Results of a systematic review. *Quality of Life Research,* 10: 347-357.

Eiser C, Vance YH & Seamark D (2000) The development of a theoretically driven measure of quality of life for children aged 6-11 years. *Child Care Health Development,* 26: 445-456.

Eley S (1997) Nutrition discussion forum – nutrition research using electronic mail. *British Journal of Nutrition,* 81: 413-414.

Elizabeth C, Suzanna S, Tim CF, Chi SL, Mital R & Bee-Wah L (1999) Pediatric asthma quality of life questionnaire: validation in children from Singapore. *Asian Pacific Journal of Allergy and Immunology,* 17 (3): 155-161.

EuroQoL Group (1990) EuroQol – a new facility for the measurement of health-related quality-of-life. *Health Policy,* 16: 199-208.

Fairfield G (1997) *Yorkshire Paediatric Intensive and High Dependency Care Study, Final Report.* Nuffield Institute for Health: Leeds.

Feeny D, Furlong W, Barr RD, Torrance GW, Rosenbaum P & Weitzman S (1992) A comprehensive multiattribute system for classifying the health status of survivors of childhood cancer. *Journal of Clinical Oncology,* 10 (6): 923-928.

Feeny D, Furlong W, Boyle M & Torrance G (1995) Multi-attribute health status classification systems: health utilities index. *PharmacoEconomics,* 7 (6): 490-502.

Feeny D, Leiper A, Barr RD, Furlong W, Torrance GW, Rosenbaum P & Weitzman S (1993) The comprehensive assessment of health status in survivors of childhood cancer: application to high-risk acute lymphoblastic leukaemia. *British Journal of Cancer,* 67: 1047-1052.

Feinstein AR (1987) *Clinimetrics,* Yale University Press: London.

Fekkes M, Theunissen NC, Brugman E, Veen S, Verrips EG, Koopman HM, Vogels T, Wit JM & Verloove-Vanhorick SP (2000) Development and psychometric evaluation of the TAPQOL: a health-related quality of life instrument for 1-5 year-old children. *Quality of Life Research,* 9 (8): 961-972.

Felder-Puig R, Frey E, Sonnleithner G, Feeny D, Gadner H, Barr RD, Furlong W & Topf R (2000) German cross-cultural adaptation of the Health Utilities Index and its application to a sample of childhood cancer survivors. *European Journal of Pediatrics,* 159: 283-288.

Feldman AB, Haley SM, & Coryell J (1990) Concurrent and construct validity of the Pediatric Evaluation of Disability Inventory, *Physical Therapy,* 70 (10): 602-610.

Field PA & Morse JM (1985) *Nursing Research: The Application of Qualitative Approaches,* Croom Helm: London.

Fiser DH (1992) Assessing the outcome of paediatric intensive care. *The Journal of Paediatrics,* 12 (1): 68-74.

Fisher M (1994) Identified needs of parents in PICU. *Critical Care Nurse,* June: 82-90.

Fitzpatrick R (2000) Measurement issues in health-related quality of life: Challenges for health psychology, *Psychology and Health,* 15: 99-108.

Fitzpatrick R, Davey C, Buxton MJ & Jones DR (1998a) Evaluating patient-based outcome measures for use in clinical trials. *Health Technology Assessment,* 2 (14).

Fitzpatrick R, Davey C, Buxton MJ & Jones DJ (1998b) Patient-assessed outcome measures. In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods – A Guide to Best Practice.* BMJ Books: London, 13-22.

Fitzpatrick R, Fletcher A, Gore S, Jones D, Spiegelhalter D & Cox D (1992) Quality of life measures in health care. I: Applications and issues in assessment. *BMJ,* 305: 1074-1077.

Fletcher A, Gore S, Jones D, Fitzpatrick R, Spiegelhalter D & Cox D (1992) Quality of life measures in health care. II: Design, analysis and interpretation. *BMJ,* 305: 1145-1148.

Fowler FJ (1993) *Survey Research Methods* (2nd Edition), Sage Publications Inc: Newbury Park.

Fowler, Jr FJ (1996) Data collection methods, In: Spilker B (Ed) *Quality of Life in Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 381-386.

Frederick CJ & Pynoos RS (1988) The Child Post-Traumatic Stress Disorder (PTSD) Reaction Index. University of California: Los Angeles.

French DJ, Carroll A & Christie MJ (1998) Health-related quality of life in Australian children with asthma: lessons for the cross-cultural use of quality of life instruments. *Quality of Life Research,* 7: 409-419.

French DJ, Christie MJ & Sowden AJ (1994a) The reproducibility of the Childhood Asthma Questionnaires: measures of quality of life for children with asthma aged 4-16 years, *Quality of Life Research*, 3: 215-224.

French DJ, Christie MJ & West A (1992) Some psychometric properties of a questionnaire measure of quality of life (QOL) in childhood asthma. *American Review Respiratory Disorders,* 145 (Suppl.): A707.

French DJ, Christie MJ & West A (1994b) Quality of life in childhood asthma: development of the Childhood Asthma Questionnaires, In: Christie M & French D (Eds) *Assessment of Quality of Life in Childhood Asthma,* Switzerland: Harwood Academic Publishers, 157-180.

Froberg DG & Kane RL (1989) Methodology for measuring health-state preferences – III: population and context effects. *Journal of Clinical Epidemiology,* 42 (6): 585-592.

Fulbrook P & Foxcroft D (1999) Measuring the outcome of paediatric intensive care. *Intensive and Critical Care Nursing,* 15: 44-51.

Garratt A (1998) (Unpublished) *Measurement of Patient Outcome.* University of York: York.

Geigle R & Jones SB (1990) Outcomes measurement: A report from the front (invited paper). *Inquiry,* 27: 7-13.

Gemke RJBJ (1999) Outcome following intensive care, In: Macnab A, Macrae D & Henning R (Eds) *Care of the Critically Ill Child,* Churchill Livingstone: London, 498-504.

Gemke RJ & Bonsel GJ (1995) Comparative assessment of paediatric intensive care. Pediatric Intensive Care Assessment of Outcome (PICASSO) Study Group. *Critical Care Medicine,* 23 (2): 238-245.

Gemke RJBJ & Bonsel GJ (1996) Reliability and validity of a comprehensive health status measure in a heterogeneous population of children admitted to intensive care. *Journal of Clinical Epidemiology,* 49 (3): 327-333.

Gemke RJBJ, Bonsel GJ & van Vught AJ (1995) Long-term survival and state of health after paediatric intensive care. *Archives of Disease in Childhood,* 73: 196-201.

Gemke RJBJ, van Vught AJ & Bonsel GJ (1993) Assessing the outcome of paediatric intensive care. *Journal of Pediatrics,* 122 (2): 325.

Gibson PG, Henry RL, Vimpani GV & Halliday J (1995) Asthma knowledge, attitudes, and quality of life in adolescents. *Archives of Diseases in Childhood,* 73 (4): 321-326.

Glaser AW, Davies K, Walker D & Brazier D (1997a) Influence of proxy respondents and mode of administration on health status assessment following central nervous system tumours in childhood. *Quality of Life Research,* 6: 43-53.

Glaser AW, Furlong W, Walker DA, Fielding K, Davies K, Feeny DH & Barr RD (1999) Applicability of the Health Utilities Index to a population of childhood survivors of central nervous system tumours in the U.K. *European Journal of Cancer,* 35 (2): 256-261.

Glaser AW, Rashid Abdul Rashid NF U CL & Walker DA (1997b) School behaviour and health status after central nervous system tumours in childhood. *British Journal of Cancer,* 76 (5): 643-650.

Gortmaker SL, Lenderking WR, Clark C, Lee S, Fowler MG, Oleske JM et al (1998) Development and use of a pediatric quality of life questionnaire in AIDS clinical trials: Reliability and validity of the general health assessment for children. In, Drotar (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents, Implications for Research and Practice,* Lawrence Erlbaum Associates: Mahwah, New Jersey, 219-236.

Gotay CC (1996) Patient-reported assessments versus performance-based tests. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 413-420.

Graham P, Stevenson J & Flynn D (1997) A new measure of health-related quality of life for children: preliminary findings. *Psychology and Health,* 12: 655-665.

Grange A (2001) *Health-related Quality of Life (HRQoL) after Paediatric Intensive Care (A Report to the NHS Northern and Yorkshire R & D Regional Office)*, LTH Hospitals NHS Trust: Leeds.

Grange A & Russell I (2000) Health-related quality of life (HRQoL) after paediatric intensive care (PIC): development and validation of a package of outcome measures. *Quality of Life Newsletter,* 24: 9.

Greenhalgh T (1997a) How to read a paper: Papers that summarise other papers (systematic reviews and meta-analyses). *BMJ,* 315: 672-675.

Greenhalgh T (1997b) How to read a paper: The Medline database. *BMJ,* 315: 180-183.

Greenhalgh J, Long AF, Brettle AJ & Grant MJ (1998) Reviewing and selecting outcome measures for use in routine practice. *Journal of Evaluation in Clinical Practice,* 4 (4): 339-350.

Greenhalgh J & Meadows K (1999) The effectiveness of the use of patient-based outcome measures of health in routine practice in improving the process and outcomes of patient care: a literature review. *Journal of Clinical Evaluation in Clinical Practice,* 5 (4): 401-416.

Grodin MA & Glantz CH (1994) *Children as Research Subjects.* Oxford University Press: New York.

Guadagnoli E & Velicer WF (1988) Relation of sample size to stability of component patters. *Psychological Bulletin,* 103: 265-275.

Gullone E & Cummins RA (1999) The comprehensive quality of life scale: A psychometric evaluation with an adolescent sample. *Behaviour Change,* 16 (2): 127-139.

Guyatt GH, Feeny DH & Patrick DL (1993) Measuring health-related quality of life. *Annals of Internal Medicine,* 118: 622-629.

Guyatt GH, Jaeschke R, Feeny DH & Patrick DL (1996) Measurements in clinical trials: choosing the right approach. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 41-48.

Guyatt GH, Kirshner B & Jaeschke R (1992) Measuring health status: What are the necessary measurement properties? *Journal of Clinical Epidemiology,* 45 (12): 1341-1345.

Guyatt G, Walter S & Norman TG (1987) Measuring change over time: assessing the usefulness of evaluative instruments. *Journal of Chronic Diseases,* 40: 171-178.

Haley SM, Coster WJ & Ludlow LH (1991) Pediatric functional outcome measures. *Physical Medicine and Rehabilitation Clinics of North America*, 2 (4): 689-723.

Haley SM, Faas RM, Coster WJ et al (1989) Pediatric Evaluation of Disability Inventory (PEDI). New England Medical Centre: Boston.

Hall A, Mcleod A, Camsell C, Thomson L & Mutch (1995) School attainment, cognitive ability and motor function in a total Scottish very-low-birthweight population at eight years: a controlled study. *Dev. Med. Child Neurology.* 37 (12): 1037-1050.

Harris H & Inayat (1997) Semi-structured interview schedules for gathering psychosocial data. *Nurse Researcher,* 5 (1): 73-85.

Hayes JA, Black NA, Jenkinson C, Young JD, Rowan KM, Daly K et al (2000) Outcome measures for adult critical care: a systematic review. *Health Technology Assessment,* 4 (24).

Hazinski MF (1992) *Nursing Care of the Critically Ill Child* (2nd Edition), Mosby Year Book: St Louis.

Heaf BP (1986) Organisation of paediatric intensive care. *Care of the Critically Ill,* 2: 141-142.

Horowitz MJ, Wilner N & Alvarez W (1979) Impact of Event Scale: A measure of subjective stress. *Psychosomatic Medicine,* 41 (3): 209-218.

Howard S, Normand C, Mugford M, Elbourne D, Field D, Johnson A, Enock K & Grant A (1995) Costing neonatal care alongside the collaborative ECMO trial: how much primary research is required? *Health Economics,* 4: 265-271.

Hutchinson JF (1996) Quality of life in ethnic groups, In: Spilker B (Ed) *Quality of Life in Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 587-593.

Hutchinson S & Skodol-Wilson H (1992) Validity threats in scheduled semi-structured interviews. *Nursing Research,* 41: 117-119.

Hutton JL & Ashcroft R (1998) What does "systematic" mean for review of methods? In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods – A Guide to Best Practice,* BMJ Books: London, 249-254.

ICNARC (1995) P*roposal for Audit of Intensive Care and High Dependency Care,* ICNARC: London.

Jack B & Clarke AM (1998) The purpose and use of questionnaires in research. *Professional Nurse,* 14 (3): 176-179.

Jacobson AM & Fried K (1998) Conceptual issues in developing quality of life assessments in children: Illustrations from studies of insulin-dependent diabetes mellitus. In, Drotar (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents, Implications for Research and Practice,* Lawrence Erlbaum Associates: Mahwah, New Jersey, 131-150.

Jellineck MS, & Murphy JM (1990) The recognition of psychological disorders in pediatric office practice: the current status of the pediatric symptom checklist. *Developmental and Behavioural Pediatrics,* 11 (5): 273-278.

Jenkinson C (1994) (Ed) *Measuring Health and Medical Outcomes,* UCL Press: London.

Jenkinson C, Layte R, Jenkinson D, et al. (1997) A shorter form health survey: can the SF12 replicate the results from the SF36 in longitudinal studies? *Journal of Public Health Medicine,* 19: 179-186.

Jenkinson C & McGee H (1997) Patient assessed outcomes: measuring health status and quality of life. In: Jenkinson C (Ed) *Assessment of Health and Health Care,* Oxford University Press: Buckingham, 64-84.

Jenkinson C & McGee H (1998) *Health Status Measurement – A Brief Critical Introduction.* Radcliffe Medical Press: Oxon.

Jenney MEM, Kane RL & Lurie N (1995) Developing a measure of health outcomes in survivors of childhood cancer: a review of the issues. *Medical Pediatric Oncology,* 24: 145-153.

Jirojanakul P & Skevington S (2000) Developing a quality of life measure for children aged 5-8 years. *British Journal of Health Psychology,* 5: 299-321.

Johnston M, Marteau T, Partridge CJ & Gilbert P (1990) Changes in patient perceptions of chronic disease and disability with time and experience. In Schmidt LR, Schwenkmezger P, Weinman J & Maes S (Eds) *Theoretical and Applied Aspects of Health Psychology*: Harwood Academic Publishers, 361-369.

Juniper EF, Guyatt GH, Feeny DH, Ferrie PJ, Griffith LE & Townsend M (1996) Measuring quality of life in children with asthma. *Quality of Life Research,* 5 (1): 35-46.

Juniper EF, Guyatt GH, Feeny DH, Griffith LE & Ferrie PJ (1997) Minimum skills required by children to complete health-related quality of life instruments for asthma: comparison of measurement properties. *European Respiratory Journal,* 10 (10): 2285-2294.

Juniper E, Guyatt GH & Jaeschke (1996) Developing a health-related quality of life measure. In: Spilker B (Ed) *Quality of Life and Pharmoeconomics in Clinical Trials,* Lippincott-Raven Publishers: Philadelphia, 49-56.

Juniper E, Guyatt G, Willan A & Griffith L (1994) Determining a minimally important change in a disease specific questionnaire. *Journal of Clinical Epidemiology,* 47: 81-87.

Kaiser HF (1958) The varimax criterion for analytic rotation in factor analysis. *Psychometrika,* 23: 187-200.

Kendrick T (2000) The impact of PICU admission on children. *Pediatric Critical Care Medicine,* 1 (1) (Supplement – 3rd World Congress on Paediatric Intensive Care June 25-29, 2000, Montréal, Canada – Book of Abstracts): 59.

Kenworthy OT, Bess FH, Stahlman MT & Lindstrom DP (1987) Hearing, speech and language outcome in infants with extreme immaturity. *American Journal of Otology,* 8 (5): 419-425.

Kessler RC & Mroczek DK (1996) Some methodological issues in the development of quality of life measures for the evaluation of medical interventions. *Journal of Evaluation in Clinical Practice,* 2 (3): 181-191.

Kinnear PR & Gray CD (1995) *SPSS for Windows Made Simple,* Lawrence Erlbaum Associates Publishers: Hove.

Kirshner B & Guyatt G (1985) A methodological framework for assessing health indices. *Journal of Chronic Disease,* 38 (1): 27-36.

Kitzinger J (1996) Introducing focus groups, In: Mays N & Pope C (Eds) *Qualitative Research in Health Care,* BMJ Publishing Group: London, 36-45.

Klem SA, Pollack MM & Getson PR (1990) Cost, resource utilization, and severity of illness in intensive care. *The Journal of Pediatrics,* 116 (2): 231-237.

Kline P (1986) *A Handbook of Test Construction,* Methuen: London.

Kline P (1994) *An Easy Guide to Factor Analysis,* Routledge: London.

Kromer ME, Prihoda TJ, Hidalgo HA & Wood PR (2000) Assessing the quality of life of Mexican-American children with asthma: impact on family function and functional status. *Journal of Pediatric Psychology,* 25 (6): 415-426.

Lakeman R (1997) Using the Internet for data collection in nursing research. *Computers in Nursing,* 15 (5): 269-275.

Landgraf JM (1999) Measuring pediatric outcomes in applied clinical settings: an update about the Child Health Questionnaire (CHQ). *Quality of Life Newsletter,* 23 (Sept-Dec).

Landgraf J (2000) http://www.healthact.com/chq/faq.htm.

Landgraf JM & Abetz LN (1996) Measuring health outcomes in paediatric populations in Spilker B. *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 793-802.

Landgraf JM & Abetz LN (1997) Functional status and well-being of children representing three cultural groups: initial self-reports using the CHQ-CF87. *Psychology and Health,* 12: 839-854.

Landgraf JM & Abetz L (1998) Influences of sociodemographic characteristics on parental reports of children's physical and psychosocial well-being: Early experiences with the Child Health Questionnaire. In, Drotar (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents, Implications for Research and Practice,* Lawrence Erlbaum Associates: Mahwah, New Jersey, 105-129.

Landgraf JM, Abetz L & Ware JE (1996) *Child Health Questionnaire (CHQ): A User's Manual. First Edition.* The Health Institute, New England Medical Center: Boston.

Landgraf JM, Abetz L, Ware JE & Maunsell E (1998) Canadian-French, German and UK versions of the Child Health Questionnaire: methodology and preliminary item scaling results. *Quality of Life Research,* 7 (5): 433-445.

Lane P (2001) Focus group methodology. *Nurse Researcher,* 8 (3): 45-56.

Law M & Usher P (1988) Validation of the Klein-Bell Activities of Daily Living Schedule for children. *Canadian Journal of Occupational Therapy,* 55: 63-68.

Lawford J, Volavka N & Eiser C (2001) A generic measure of Quality of Life for children aged 3-8 years: results of two preliminary studies. *Pediatric Rehabilitation,* 4 (4): 197-207.

Le Coq EM, Boeke AJ, Bezemer PD, Colland VT & van Eijk JT (2000a) Which source should we use to measure quality of life in children with asthma: the children themselves or their parents? *Quality of Life Research,* 9 (6): 625-636.

Le Coq EM, Boeke AJ, Bezemer PD, Bruil J & van Eijk JT (2000b) Clinimetric properties of a parent report on their offspring's quality of life. *Journal of Clinical Epidemiology,* 53 (2): 139-146.

Le Coq EM, Colland VT, Boeke AJP, Bezemer DP & van Eijk TH (2000c) Reproducibility, construct validity, and responsiveness of the "How Are You" (HAY), a self-report quality of life questionnaire for children with asthma. *Journal of Asthma,* 37 91): 43-58.

Le Gales C, Costet N, Gentet JC, Kalifa C, Frappez D, Edan C, Sariban E, Plantaz D & Doz F (1999) Cross-cultural adaptation of a health status classification system in children with cancer. First results of the French adaptation of the Health Utilities Index Marks 2 and 3. *International Journal of Cancer,* (Supplement 12): S112-118.

Leventhal H, Meyer D & Nerenz (1980) The common sense representation of illness danger. In: Rachman S (Ed) *Medical Psychology (Volume 2)*, Pergamon: New York, 7-30.

Leventhal H & Nerenz D (1985) The assessment of illness cognition. In: Skelton JA & Croyle RT (Eds) *Measurement in Health Psychology,* Wiley: New York, 517-555.

Leventhal H, Nerenz DR & Steele DJ (1984) Illness representations and coping with health threats. In Baum A, Taylor SE & Singer JE (Eds) *Handbook of Psychology and Health, Volume IV, Social Psychological Aspects of Health,* Wiley: New York, 517-555.

Levi R & Drotar D (1998) Critical issues and needs in health-related quality of life assessment of children and adolescents with chronic health conditions. In: Drotar D (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents, Implications for Research and Practice,* Lawrence Erlbaum Associates: New Jersey, 3-24.

Lindström B & Köhler L (1991) Youth, disability and quality of life. *Pediatrician,* 18: 121-128.

Lindström B & Eriksson B (1993) Quality of life among children in the Nordic countries. *Quality of Life Research,* 2: 23-32.

Lohr KN (1988) Outcomes measurement: concepts and questions. *Inquiry,* 25: 37-50.

Lowe HJ & Barnett GO (1994) Understanding and using the Medical Subject Headings (MeSH) Vocabulary to perform literature searches. *Journal of the American Medical Association,* 271: 1103-1108.

MacKeigan LD & Pathak DS (1992) Overview of health-related quality of life measures. *American Journal of Hospital Pharmacy,* 49: 2236-2245.

Madagame ET, Havens PL, Bresnahan JM, Babel KL & Splaingard ML (1995) Survival and functional outcome of children requiring mechanical ventilation during therapy for acute bacterial meningitis. *Critical Care Medicine*, 23 (7): 1279-1283.

Magaziner J, Simonsick E, Kashner M & Hebel J (1988) Patient-proxy response comparability on measures of patient health and functional status. *Journal of Clinical Epidemiology,* 41: 1065-1074.

Manificat S & Dazord A (1998) Children's quality of life assessment: preliminary results obtained with the AUQEI questionnaire, *Quality of Life Newsletter,* 19 (Jan-April).

Manificat S, Dazord A, Langue J, Danjou G, Bauche P, Bovet F, Cubells JM, Luchelli R, Tockert E & Conway K (1999) A new instrument to evaluate infant quality of life, *Quality of Life Newsletter,* 23 (Sept-Dec).

Manificat S, Dazord A, Nicolas J, Cochat P & David L (1995) Quality of life evaluation with children: validation of a questionnaire. *Quality of Life Newsletter,* 12 (Feb-May).

Marra C, Levine M, McKerrow R & Carleton B (1996) Overview of health-related quality-of-life measures for pediatric patients: Application in the assessment of pharmacotherapeutic and pharmacoeconomic outcomes. *Pharmacotherapy,* 16 (5): 879-888.

Marteau TM & Bekker H (1992) The development of a six-item short-form of the state scale of the Spielberger State-Trait Anxiety Inventory (STAI). *British Journal of Clinical Psychology,* 31: 301-306.

Marteau T & Johnston M (1986) Determinants of beliefs about illness: a study of parents and children with diabetes, asthma, epilepsy, and no chronic illness. *Journal of Psychosomatic Research,* 30 (6): 673-683.

Mason J (1996) *Qualitative Researching,* Sage Publications Ltd: London.

Mays N & Pope C (Eds) (1996a) *Qualitative Research in Health Care,* BMJ Publishing Group: London.

Mays C & Pope N (1996b) Rigour and qualitative research. In Mays C & Pope N (Eds) *Qualitative Research in Health Care,* BMJ Publishing Group: London, 10-19.

McAloon J, Crean P, Jenkins J & McGlure G (1991) Evaluation of paediatric intensive care in a regional centre. *Archives of Disease in Childhood,* 66: 1043-1046.

McColl E, Jacoby A, Thomas L, Soutter J, Bamford C, Garratt A, Harvey E, Thomas R & Bond J (1998) Designing and using patient and staff questionnaires, In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods – A Guide to Best Practice*, BMJ Books: London, 46-58.

McColl E, Jacoby A, Thomas L, Soutter J, Bamford C, Steen N et al, (2001) Design and use of questionnaires: a review of best practice applicable to surveys of health service staff and patients. *Health Technology Assessment,* 5 (31).

McDowell I & Jenkinson C (1996) Development standards for health measures, *Journal of Health Services Research & Policy,* 1 (4): 238-246.

M°Dowell I & Newell C (1996) *Measuring Health: A Guide to Rating Scales and Questionnaires.* Oxford University Press: New York.

McGee HM, O'Boyle CA, Hickey A, O'Malley K & Boyce CRB (1991) Assessing the quality of life of the individual: the SEIQoL with a healthy and a gastroenterology unit population. *Psychological Medicine,* 21: 749-759.

McGrath MM, Bukstein DA, Buchner DA, Guzman GI, Landgraf JM & Goss TF (1999) Assessment of the relationship between disease severity and general and disease-specific health-related quality of life in pediatric asthma patients. *Ambulatory Child Health,* 5 (3): 249-261.

Miles MB & Huberman AM (1984) *Qualitative Data Analysis: A Sourcebook of New Methods,* Sage Publications Inc: London.

Mishoe SC, Baker RR, Poole S, Harrell LM, Avant CB & Rupp NT (1998) Development of an instrument to assess stress levels and quality of life in children with asthma. *Journal of Asthma,* 35 (7): 553-563.

Mok Q & Butt W (1996) The outcome of children admitted to intensive care with meningococcal septicaemia. *Intensive Care Medicine,* 22: 259-263.

Morrison A, Mellis C, O'Connell A, Schell D & Gillis J (2000) Outcome of pediatric intensive care. *Pediatric Critical Care Medicine,* 1 (1) (Supplement – 3rd World Congress on Paediatric Intensive Care June 25-29, 2000, Montréal, Canada – Book of Abstracts): 60.

Morse J (Ed) (1991) *Qualitative Nursing Research,* Sage Publications Inc: London.

Moseley LG, Mead DM & Murphy FA (1997) Applying lexical and semantic analysis to the exploration of free-text data. *Nurse Researcher,* 4 (3): 46-68.

Moser CA & Kalton G (1996) *Survey Methods in Social Investigation,* Dartmouth: Aldershot.

Msall ME, DiGaudio KM & Duffy LC (1993) Use of functional assessment in children with developmental disabilities. *Physical Medicine & Rehabilitation Clinics of North America,* 4 (3): 517-527.

Mulhern RK (1999) Correlation of the Health Utilities Index Mark 2 cognition scale and neuropsychological functioning among survivors of childhood medulloblastoma. *International Journal Of Cancer,* Supplement 12: 91-94.

Munzenberger PJ, van Wagnen CA, Abdulhamid I & Walker PC (1999) Quality of life as a treatment outcome in patients with cystic fibrosis. *Pharmacotherapy,* 19 (4): 393-398.

Murray PJ (1995) Research data from cyberspace: interviewing nurses by e-mail. *Health Informatics,* 1 (2): 73-76.

National Co-ordinating Centre for HTA (1997) *Identifying Questions, Finding Answers. Annual Report NHS Health Technology Assessment Programme 1997,* Leeds: NHS Executive.

Nelson EC, Landgraf JM, Hays RD, Wasson JH & Kirk JW (1990) The functional status of patients: how can it be measured in physician's offices? *Medical Care,* 28 (12): 1111-1126.

Nelson EC, Wasson JH, Johnson DJ & Hays RD (1996) *Dartmouth COOP Functional Health Assessment Charts: Brief Measures for Clinical Practice* In Spilker B (Ed) Quality of Life and Pharmocoeconomics in Clinical Trials (2nd Edition), Lippincott-Raven Publishers: Philadelphia.

Nelson EC, Wasson J, Kirk J et al, (1987) Assessment of function in routine clinical practice: description of the COOP chart method and preliminary findings. *Journal of Chronic Diseases,* 40 (S1): 55S-63S.

Newacheck PW & Taylor WR (1992) Childhood chronic illness: prevalence, severity and impact. *American Journal of Public Health,* 82: 364-371.

NHS CRD (1996) *Undertaking Systematic Reviews of Research on Effectiveness – CRD Guidelines for Those Carrying Out or Commissioning Reviews, CRD Report 4,* NHS CRD: York.

NHSE (1995) *Northern and Yorkshire RHA Lifestyle Survey Covering the Former Yorkshire Region,* NHSE Northern & Yorkshire: Leeds.

NHSE (1996a) *Promoting Clinical Effectiveness,* Department of Health: London.

NHSE (1996b) *Paediatric Intensive Care: The Way Forward. NHS Executive Letter EL (96) 53,* Department of Health: London.

NHSE (1996c) *Paediatric Intensive Care: Report from the Chief Executive of the NHS Executive to the Secretary of State,* Department of Health: London

NHSE (1996d) *Research and Development in the Department of Health and National Health Service,* Department of Health: London.

NHSE (1997) *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.

Nord E (1991) EuroQol: health-related quality of life measurement. Valuations of health states by the general public in Norway. *Health Policy,* 18: 25-36.

Noyes J (2000) The views and experiences of young 'ventilator-dependent' people who spent prolonged periods of time in intensive care units (ICUs) when they no longer have a medical need or want to be there. *Pediatric Critical Care Medicine,* 1 (1) (Supplement – 3rd World Congress on Paediatric Intensive Care June 25-29, 2000, Montréal, Canada – Book of Abstracts): 60-61.

NRR (2000) [database available on-line] NHS Executive: London.

Nunnally JC Jr (1970) *Introduction to Psychological Measurement*, McGraw-Hill Book Company: New York.

Nunnally JC (1978) *Psychometric Theory* (2nd Edition), McGraw-Hill Book Company: New York.

O'Boyle CA, McGee H & Joyce CRB (1994) Quality of life: Assessing the individual. *Advances in Medical Sociology,* 5: 159-180.

Oppenheim AN (1992) *Questionnaire Design, Interviewing and Attitude Measurement* (New Edition), Pinter Publishers: London.

Orenstein DM, Nixon PA, Ross EA & Kaplan RM (1989) The quality of wellbeing in cystic fibrosis. *Chest,* 95 (2): 344-377.

Pal DK (1996) Quality of life assessment in children: a review of conceptual and methodological issues in multidimensional health status measures. *Journal of Community Health,* 50: 391-396.

Pantell RH & Lewis CC (1987) Measuring the impact of medical care on children. *Journal of Chronic Disease,* 40 (supplement 1): 99S-108S.

Parry G [Scottish Neonatal Consultants Collaborative Study Group and the International Network] (1995) CRIB, mortality and impairment after neonatal intensive care. *Lancet,* 345: 1020-1022.

Pateman B (1998) Computer-aided qualitative data analysis: the value of NUD*IST and other programs. *Nurse Researcher,* 5 (3): 77-89.

Patrick DL & Bergner M (1990) Measurement of health status in the 1990's. *Annual Review of Public Health,* 11: 165-183.

Patrick DL & Erickson P (1996) Applications of health status assessment to health policy. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 717-727.

Phagoo SB, Wilson NM & Silverman M (1995) Evaluation of a new interrupter device for measuring change in airway resistance in 5-year-old asthmatic children. *Pediatric Pulmonology,* 20 (6): 387-395.

Phagoo SB, Wilson NM & Silverman M (1996) Evaluation of a new interrupter device for measuring bronchial responsiveness and the response to bronchodilator in 3 year old children. *European Respiratory Journal,* 9 (7): 1374-1380.

Pham B & Klaassen (2000) Assessing clinical measures and clinical disagreement. In: Moyer VA (Ed) *Evidence-based Pediatrics and Child Health,* London: BMJ Books, 71-78.

PICS (1996) *Paediatric Intensive Care Society Standards for Paediatric Intensive Care,* Saldatore Ltd: Bishop's Stortford.

PICS (2001) *Paediatric Intensive Care Society Standards Document 2001*, PICS: London.

Playfor S, Thomas D & Choonara I (2000) Recollection of children following intensive care. *Archives of Diseases in Childhood,* 83: 445-448.

Polgar S & Thomas SA (1998) *Introduction to Research in the Health Sciences* (3rd Edition), Melbourne: Churchill Livingstone.

Pollack MM, Alexander SR, Clarke N, Ruttimann UE, Tesselaar HM & Bachulis AC (1991) Improved outcomes from tertiary center pediatric intensive care: A statewide comparison of tertiary and nontertiary care facilities. *Critical Care Medicine,* 19 (2): 150-159.

Pollack MM, Cuerdson TC & Getson PR (1993) Pediatric intensive care unit: results of a national survey. *Critical Care Medicine,* 21 (4): 607-614.

Pollack MM, Katz RW, Ruttiman UE & Getson P (1988) Improving the outcome and efficiency of intensive care: The impact of an intensivist. *Critical Care Medicine,* 16 (1): 11-17.

Pollack MM, Patel KM & Ruttiman UE (1996) PRISM III: an updated Pediatric Risk of Mortality score. *Critical Care Medicine,* 24 (5): 743-752.

Pollack MM, Ruttiman UE & Getson PR (1988) Pediatric risk of mortality (PRISM) score. *Critical Care Medicine,* 16 (11): 1110-1116.

Pollack MM, Wilkinson JD & Glass N (1987) Long-stay pediatric intensive care unit patients: Outcome and resource utilisation. *Pediatrics,* 80 (6): 855-860.

Pope C & Mays N (1996) Qualitative methods in health and health services research, In: Mays N & Pope C (Eds) *Qualitative Research in Health Care,* BMJ Publishing Group: London, 1-9.

Prescott RJ, Gillespie WJ, Counsell CE, Grant AM, Ross S, Russell IT & Kiauka S (1998) Factors that limit the number, quality, and progress of randomised trials, In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods. A Guide to Best Practice,* BMJ Books: London, 86-97.

QSR Pty Ltd (1997) *User Guide QSR NUD.IST 4 Software for Qualitative Data Analysis* (2nd Edition), Qualitative Solutions & Research Pty Ltd: Australia.

Randolph AG (1997) Paediatric Index of Mortality (PIM): do we need another paediatric mortality prediction score? *Intensive Care Medicine,* 23; 141-142.

Raphael D, Rukholm E, Brown I, Hill-Bailey P & Donato E (1996) The Quality of Life Profile – Adolescent Version: Background, description and initial validation. *Journal of Adolescent Health,* 19: 366-375.

Ravens-Sieberer U & Bullinger M (1997) Current approaches to measure the health-related quality of life in children. *Quality of Life Newsletter,* 18: 1-3

Ravens-Sieberer U & Bullinger M (1998) Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. *Quality of Life Research,* 7: 399-407.

Reference Manager [computer program] Windows version 8.0 (1997) Research Information Systems: Carlsbad.

Reichenberg K & Broberg AG (2000) Quality of life in childhood asthma: use of the Paediatric Asthma Quality of Life Questionnaire in a Swedish sample of children 7 to 9 years old. *Acta Paediatrica,* 89 (8): 989-995.

Rennick J (1986) Re-establishing the parental role in a pediatric intensive care unit. *Journal of Pediatric Nursing,* 1: 40-45.

Rennick JE & Johnston CC (2000) Development of a measure to examine post-traumatic stress responses in children following critical illness. *Pediatric Critical Care Medicine,* 1 (1) (Supplement – 3rd World Congress on Paediatric Intensive Care June 25-29, 2000, Montréal, Canada – Book of Abstracts): 58.

Rennick JE, Johnston CC, Dougherty G, Platt R & Ritchie J (2000) Children's psychological responses following pediatric intensive care unit (PICU) hospitalization. *Pediatric Critical Care Medicine,* 1 (1) (Supplement – 3rd World Congress on Paediatric Intensive Care June 25-29, 2000, Montréal, Canada – Book of Abstracts): 58.

Riddle II, Hennessey J, Eberly TW, Carter MC & Miles MS (1989) Stressors in the paediatric intensive care unit as perceived by mothers and fathers. *Maternal Child Nursing,* 18 (2): 221-234.

Riley AW, Green BF, Forrest CB, Starfield B, Kang M & Ensminger ME (1998) A taxonomy of adolescent health: development of the adolescent health profile types. *Medical Care,* 36 (8): 1228-1236.

Roberts TE & The Extracorporeal Membrane Oxygenation Economics Working Group (1998) Economic evaluation and randomised controlled trial of extracorporeal membrane oxygenation: UK collaborative trial. *BMJ,* 317: 911-915.

Robson C (1995) R*eal World Research: A Resource for Social Scientists and Practitioner Researchers,* Blackwell: Oxford.

Rosenbaum PL & Saigal S (1996) Measuring health-related quality of life in pediatric populations: conceptual issues. In, Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 785-791.

Rosenbaum PL, Cadman D & Kripalami H (1990) Pediatrics: assessing quality of life. In Spilker B (Ed) *Quality of Life Assessment in Clinical Trials*, Raven Press: New York, 205.

Rothman ML, Hedrick SC, Bulcroft KA, Hickam DH & Rubenstein LZ (1984) The validity of proxy-generated scores as measures of patient health status. *Medical Care,* 22: 789-795.

Russell I (1996) What is Research and Development? *Refocus - Northern and Yorkshire R & D Newsletter,* Spring: 2a (i) – 2a (iv).

Russell I (1998) *Teaching Notes for Survey Research Methods Module* (MSc in Health Sciences), Department of Health Sciences, University of York: York (unpublished).

Russell I, Di Blasi Z, Lambert M & Russell D (1998) Systematic reviews and meta-analyses: opportunities and threats. *In:* Templeton A, O'Brien P (Eds) *Evidence-based Fertility Treatment,* RCOG Press: London, Chapter 2.

Ruta DA, Garratt AM, Leng M, Russell IT & MacDonald LM (1994) A new approach to the measurement of quality of life. The Patient-Generated Index. *Medical Care,* 32 (11): 1109-1126.

Saigal S, Feeny D, Furlong W, Rosenbaum P, Burrows E & Torrance G (1994a) Comparison of the health-related quality of life of extremely low birth weight children and a reference group of children at age eight years. *Journal of Pediatrics,* 125 (3): 418-425.

Saigal S, Rosenbaum P, Hoult L, Furlong W, Feeny D, Burrows E & Stoskopf B (1998a) Conceptual and methodological issues in assessing health-related quality of life in children and adolescents: illustration from studies of extremely low birthweight survivors. In, Drotar (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents, Implications for Research and Practice,* Lawrence Erlbaum Associates: Mahwah, New Jersey, 151-169.

Saigal S, Rosenbaum P, Stoskopf B, Hoult L, Furlong W, Feeny D, Burrows E & Torrance G (1994b) Comprehensive assessment of the health status of extremely low birth weight children at eight years of age: comparison with a reference group. *The Journal of Pediatrics,* 125 (3): 411-417.

Saigal S, Stoskopf BL, Rosenbaum PL, Hoult LA, Furlong WJ & Feeny DH (1998b) Development of a multi-attribute pre-school health status classification system. *Pediatric Research,* 43: 228 (abstract).

Sapsford R & Abbott P (1992) R*esearch Methods for Nurses and the Caring Professions,* Open University Press: Bristol.

Scharloo M, Kaptein AA, Weinman J, Bergman W, Vermeer BJ & Rooijmans HG (2000a) Patients' illness perceptions and coping as predictors of functional status in psoriasis: a 1-year follow-up. *British Journal of Dermatology,* 142 (5): 899-907.

Scharloo M, Kaptein AA, Weinman JA, Willems LN & Rooijmans HG (2000b) Physical and psychological correlates of functioning in patients with chronic obstructive pulmonary disease. *Journal of Asthma,* 37 (1): 17-29.

Schipper H, Clinch JJ & Olweny CLM (1996) Quality of life studies: Definitions and conceptual issues. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 11-23.

Scholle SH, Whiteside L, Kelleher K, Bradley R & Casey P (1995) Health status of preterm low-birth-weight infants. *Archive of Pediatric and Adolescent Medicine,* 149: 1351-1357.

Schor EL (1995) The influence of families on child health. *Pediatric Clinics of North America,* 42: 89-101.

Schor EL (1998) Children's health and assessment of health-related quality of life. In. Drotar D (Ed) *Measuring Health-Related Quality of Life in Children and Adolescents, Implications for Research and Practice,* Lawrence Erlbaum Associates: Mahwah, New Jersey, 25-37.

Schor EL, Lerner DJ & Malspeis S (1995) Physicians' assessment of functional status and wellbeing. *Archives Internal Medicine,* 155, 309-314.

Schwandt T (1994) Constructivist, Interpretist Persuasions for Human Inquiry. In: Denzin N & Lincoln Y (Eds) *Handbook of Qualitative Research,* Sage Publications Inc: Thousand Oaks, California, 118-137.

Scothern G et al (1992) Young children in a general intensive therapy unit. *Care of the Critically Ill,* 8 (5): 208-209.

Seid M, Varni JW & Kurtin PS (2000) Measuring quality of care for vulnerable children: challenges and conceptualisation of a pediatric outcome measure of quality. *American Journal of Medical Quality,* 15 (4): 182-188.

Seid M, Varni JW, Rode CA & Katz ER (1999a) The Pediatric Cancer Quality of Life inventory: a modular approach to measuring health-related quality of life in children with cancer. *International Journal of Cancer* (12 Supplement): S71-S76.

Seid M, Varni JW & Seizer IS (1999b) The PedsQL: a modular approach to measuring health-related quality of life in children with arthritis, EULA VIth International Pediatric Rheumatology Conference, Glasgow, Scotland, June 6-11th . *Ann. Rheum. Dis.* 352: 1490.

Segedin E (1999) Epidemiology of severe illness in childhood. In: McNab A, Macrae D J & Henning R (Eds) *Care of the Critically Ill Child,* Churchill Livingstone: London, 12-23.

Sell RL (1997) Research and the Internet: an e-mail survey of sexual orientation. *American Journal of Public Health,* 87 (2): 297.

Shaffer D, Gould M, Brasic J et al (1983) A children's global assessment scale (CGAS). *Archives of General Psychiatry,* 40: 1228-1231.

Shann F, Pearson G, Slater A & Wilkinson K (1997) Paediatric Index of Mortality (PIM): a mortality prediction model for children in intensive care. *Intensive Care Medicine,* 23 (2): 201-207.

Shumaker SA, Anderson RT & Czajkowski SM (1990) Psychological tests and scales. In Spilker B (Ed) *Quality of Life Assessments in Clinical Trials,* New York: Raven Press. 95-113.

Simeoni MC, Auquier P, Antoniotti S, Sapin C & San Marco JL (2000) Validation of a French health-related quality of life instrument for adolescents: The VSP-A. *Quality of Life Research,* 9: 393-403.

Sinclair J (1997) The spectrum of paediatric intensive care In: Morton NS (Ed*) Paediatric Intensive Care*, Oxford University Press: Oxford, 1-30.

Singh-Naz N, Sprague BM, Patel KM & Pollack MM (1996) Risk factors for nosocomial infection in critically ill children: A prospective cohort study. *Critical Care Medicine,* 24 (5): 875-878.

Speechley KN, Maunsell E, Desmeules M, Schanzer D, Landgraf JM, Feeny DH & Barrera ME (1999) Mutual concurrent validity of the Child Health Questionnaire and the Health Utilities Index: An exploratory analysis using survivors of childhood cancer. *International Journal of Cancer,* (Supplement 12): S95-S105.

Speith LE & Harris CV (1996) Assessment of health-related quality of life in children and adolescents: An integrative review. *Journal of Pediatric Psychology,* 21: 175-193.

Spencer NJ & Coe C (1996) The development and validation of a measure of parent-reported child-health and morbidity: The Warwick Child Health and Morbidity Profile. *Child: Care, Health and Development,* 22 (6): 367-379.

Spielberger CD (1983) *Manual for the State-Trait Anxiety Inventory STAI (Form Y),* Palo Alto, CA: Consulting Psychologists Press.

Spielberger CD, Gorsuch RL & Lushene RE (1970) *Manual for the State-Trait Anxiety Inventory.* Palo Alto, CA: Consulting Psychologists Press.

Spilker B (1996) Introduction In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials (2nd Edition),* Lippincott-Raven Publishers: Philadelphia, 1-10.

Spilker B & Revicki A (1996) Taxonomy of quality of life, In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 25-31.

Sprangers MAG & Aarson NK (1992) The role of health care providers and significant others in evaluating quality of life of patients with chronic disease: a review. *Journal of Clinical Epidemiology,* 45 (7): 743-760.

SPSS Inc. (1999) *SPSS Release 10 for Windows,* SPSS Inc. Chicago.

Starfield B (1987) Child health status and outcome of care: a commentary on measuring the impact of medical care on children. *Journal of Chronic Diseases,* 40 (Supplement 1): 109S – 115S.

Starfield B, Bergner M, Ensminger M, Riley A, Ryan S, Green B, McGauhey P, Skinner A & Kim S (1993) Adolescent Health Status Measurement: Development of the Child Health and Illness Profile. *Pediatrics,* 91(2): 430-435.

Starfield B, Riley A, Green B, Ensminger ME, Ryan SA, Kelleher K, Kim-Harris S, Johnston D & Vogel K (1995) The Adolescent Child Health & Illness Profile – A Population-Based Measure of health. *Medical Care* 33(5): 553-566.

Stein REK & Jessop DJ (1982) A noncategorical approach to chronic childhood illness. *Public Health Reports,* 97 (4): 354-362.

Stein REK & Jessop DJ (1990) Functional II (R) – A Measure of Child Health Status [erratum appears in *Medical Care* (1991), May, 29 (5): following 489]. *Medical Care,* 28(11): 1041-1055.

Steinwachs DM, Wu AW & Cagney KA (1996) Outcomes research and quality of care. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials (2nd Edition),* Lippincott-Raven Publishers: Philadelphia, 747-752.

Stern PN (1991) Are counting and coding a cappella appropriate in qualitative research? In: Morse J (Ed) *Qualitative Nursing Research,* Sage Publications Inc: London, 147-161.

Stevens VG, Hibbert CL & Edbrooke DL (1998) Evaluation of proposed case-mix criteria as a basis for costing patients in the adult general intensive care unit. *Anaesthesia,* 53: 944-950.

Stevens A, Milne R & Black N (1998) Researching Health Services, In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods – A Guide to Best Practice*, BMJ Books: London, 1-9.

Stevenson RC, M^{c}Cabe CJ, Pharoah PO, Cooke RW (1996a) Cost of care for a geographically determined population of low birthweight infants to age 8-9 years I. Children with disability. *Archives of Diseases in Childhood.* 74 (2): F114-117.

Stevenson RC, Pharoah PO, Stevenson CJ, M^{c}Cabe CJ, Cooke RW (1996b) Cost of care for a geographically determined population of low birthweight infants to age 8-9 years II. Children with disability. *Archives of Diseases in Childhood.* 74 (2): F118-121.

Stewart AL, Greenfield S, Hays RD, Wells K, Rogers WH, Berry SD, M^{c}Glynn EA & Ware JE (1989) Functional status and well-being of patients with chronic conditions: results from the medical outcomes study. *Journal of the American Medical Association,* 262: 907-913.

St Leger AS, Schnieden H & Walsworth-Bell JP (1994) *Evaluating Health Services' Effectiveness. A Guide for Health Professionals, Service Managers and Policy Makers,* Open University Press: Milton Keynes.

Stolk EA, Busschbach JJ & Vogels T (2000) Performance of the Euroqol in children with imperforate anus. *Quality of Life Research,* 9 (1): 29-38.

Streiner D L & Norman G R (1995) *Health Measurement Scales: A Practical Guide to Their Development and Use* (2nd Edition), Oxford University Press: Oxford.

Sutton AJ, Jones DR, Abrams K, Sheldon TA & Song F (1998) Systematic reviews of randomised trials In: Black N, Brazier J, Fitzpatrick R & Reeves B (Eds) *Health Services Research Methods – A Guide to Best Practice,* BMJ Books: London, 175-186.

Szecket N, Medin G, Furlong WJ, Feeny DH, Barr DH & Depauw S (1999) Preliminary translation and cultural adaptation of Health Utilities Index Questionnaires for application in Argentina. *International Journal of Cancer,* (Supplement 12): S119-S124.

Tabachnick BG & Fidell LS (2001) *Using Multivariate Statistics* (4th Edition), Allyn and Bacon: Boston.

Taylor WR & Newacheck P (1992) Impact of childhood asthma on health. *Pediatrics,* 90 (5): 657-662.

Teres D & Lieberman S (1991) Are we ready to regionalize pediatric intensive care? *Critical Care Medicine,* 19 (2): 139-140.

Theunisson NCM, Vogels TGC, Koopman HM, Verrips GHW, Zwinderman KAH, Verloove-Vanhorick SP & Wit JM (1998) The proxy problem: children report versus parent report in health-related quality of life research. *Quality of Life Research,* 7: 387-397.

Titman P, Smith M & Graham P (1997) Assessment of the quality of life of children. *Clinical Child Psychology and Psychiatry,* 2 (4): 597-606.

Torrance GW, Feeny DH, Furlong WJ, Barr RD, Zhang Y & Wang Q (1996) Multiattribute utility function for a comprehensive health status classification system – Health Utilities Index Mark 2. *Medical Care,* 34 (7): 702-722.

Torrance GW, Furlong W, Feeny D & Boyle M (1995) Multi-attribute preference functions – Health Utilities Index. *PharmacoEconomics* 7 (6): 503-520.

Trudel JG, Rivard M, Dobkin PL, Leclerc J-M & Robaey P (1998) Psychometric properties of the Health Utilities Index Mark 2 system in paediatric oncology patients. *Quality of Life Research,* 7: 421-432.

UK Collaborative ECMO Trial Group (1996) UK collaborative randomised trial of neonatal extracorporeal membrane oxygenation. *Lancet,* 348: 75-82.

Varni JW (1998) http:www.pedsql.org

Varni JW, Katz ER, Seid M, Quiggins DJL, Friedman-Bender A & Castro CM (1998a) The Pediatric Cancer Quality of Life Inventory (PCQL): I, Instrument development, descriptive statistics, and cross-informant variance. *Journal of Behavioural Medicine,* 21: 179.

Varni JW, Katz ER, Seid M, Quiggins DJL, Friedman-Bender A (1998b) The Pediatric Cancer Quality of Life Inventory-32 (PCQL-32): I, Reliability and validity. *Cancer,* 82; 1184.

Varni JW, Rode CA, Seid M, Katz ER, Friedman-Bender A & Quiggins DJL (1999a) The Pediatric Cancer Quality of Life Inventory-32 (PCQL-32). II. Feasibility and range of measurement. *Journal of Behavioural Medicine,* 22 (4): 397-406.

Varni JW, Seid M & Kurtin PS (1999b) The PedsQL™ 4.0 Measurement model for the Pediatric Quality of Life Inventory Version 4.0 *Quality of Life Newsletter,* 23 (Sept-Dec 1999).

Varni JW, Seid M & Kurtin PS (1999c) Pediatric health-related quality of life measurement technology: A guide for health care decision makers. *JCOM,* 6 (4): 33-39.

Varni JW, Seid M & Kurtin PS (2001) PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. *Medical Care,* 39 (8): 800-812.

Varni JW, Seid M & Rode CA (1999d) The PedsQL™: Measurement model for the Pediatric Quality of Life Inventory, *Medical Care,* 37 (2): 126-139.

Verrips GHW, Vogels TGC, Fekkes M, Theunissen NCM, Koopman HM, Kamphius RP, Wit JM, Verloove-Vanhorick SP (1997) The validity of the TACQOL-child form: A quality of life questionnaire for children. *Quality of Life Research,* 6 (5): 35.

Verrips GHW, Vogels TGC, Koopman HM, Theunissen NCM, Kamphuis RP, Fekkes M, Wit JM & Verloove-Vanhorick SP (1999) Measuring health-related quality of life in a child population. *European Journal of Public Health,* 9 (3): 188-193.

Vivier PM, Bernier JA & Starfield B (1994) Current approaches to measuring health outcomes in pediatric research. *Current Opinion in Pediatrics,* 6: 530-537.

Vogels T, Verrips GHW, Verloove-Vanhorick SP, Fekkes M, Kamphuis RP, Koopman HM, Theunissen NCM & Wit J M (1998) Measuring health-related quality of life in children: the development of the TACQOL parent form. *Quality of Life Research,* 7: 457-465.

Walker LS & Greene JW (1991) The Functional Disability Inventory: measuring a neglected dimension of child health status. *Journal of Pediatric Psychology,* 16 (1): 39-58.

Ware JE (1984) Conceptualising disease impact and treatment outcomes. *Cancer,* 15 (S): 2316-2323.

Ware J (1987) Standards for validating health measures: Definition and content. *Journal of Chronic Disease,* 40 (6): 473-480.

Ware JE, Nelson EC, Sherbourne CD & Stewart AL (1992) Preliminary tests of a 6-item general health survey: A patient application. In: Stewart AL & Ware JE (Eds) *Measuring Functioning and Well-Being: The Medical Outcomes Study Approach.* Duke University Press: 291-308.

Ware JE & Sherbourne CD (1992) The MOS 36-item short form health survey (SF-36): I. Conceptual framework and item selection. *Medical Care,* 30: 473-483.

Waters E, Wright M, Wake M, Landgraf J & Salmon L (1999) Measuring the health and well-being of children and adolescents: a preliminary comparative evaluation of the Child Health Questionnaire in Australia. *Ambulatory Child Health,* 5 (2): 131-141.

Waters E, Salmon L & Wake M (2000) The parent-form child health questionnaire in Australia: Comparison of reliability, validity, structure, and norms. *Journal of Pediatric Psychology,* 25 (6): 381-391.

Waters E, Salmon L, Wake M, Hesketh K & Wright M (2000) The Child Health Questionnaire in Australia: reliability, validity and population means. *Australian and New Zealand Journal of Public Health,* 24 (2): 207-210.

Weinman J, Petrie KJ, Moss-Morris R & Horne R (1996) The Illness Perception Questionnaire: A new method for assessing the cognitive representation of illness. *Psychology and Health,* 11: 431-445.

WHO (1948) *Constitution of the World Health Organisation,* WHO: Geneva.

WHOQOL Group (1993) *Measuring Quality of Life: The Development of the World Health Organisation Quality of Life Instrument (WHOQOL),* WHO: Geneva.

Williams AH (1996) Health policy implications of using quality of life measures in the economic evaluation of health care. In: Spilker B (Ed) *Quality of Life and Pharmacoeconomics in Clinical Trials* (2nd Edition), Lippincott-Raven Publishers: Philadelphia, 753-759.

Yeh TS, Pollack MM, Holbrook PR, Fields AI & Ruttiman U (1982) assessment of pediatric intensive care – application of the therapeutic Intervention Scoring System. *Critical Care Medicine,* 10 (8): 497-500.

Yin R (1994) *Case Study Research. Design and Methods (2nd Edition),* Sage Publications Inc: London.

Zigmond AS & Snaith RP (1983) The Hospital Anxiety and Depression Scale. *Acta Psychiatrica Scandinavica,* 67: 361-370.