

**Decision Processes in the Use of Technological
Support for Children and Young People
with Life-Limiting Conditions**

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ABSTRACT

Background: Decisions about technological support for children with life-limiting conditions are surrounded by uncertainty, and require families to weigh up complex issues around quality and prolongation of life. The limited evidence suggests there is inadequate information and support for decision-making, and potential for misunderstanding between families and professionals.

Aim: To investigate parents' and young people's experiences of making decisions about technological support, focusing on artificial nutrition and assisted ventilation.

Methods: A purposive sample of nineteen families of life-limited children and young people (twenty-five parents and five young people) who had considered the use of technological support was drawn from users of a children's hospice. In-depth interviews were carried out with this sample and the data subject to thematic analysis.

Findings: Parents and young people wish to make the right choices, and consider a range of factors and information in an effort to become informed, even when there is no perceived choice in decisions they make. Quality of life is identified as the key decision factor, which as a fluid and evolving concept underpins the process of assessing a child's needs and considering the potential impact of a proposed intervention. Wider features of the process also influence how families make decisions, and the research proposes an ecological framework which distinguishes between decision factors, used by families to consider a proposed intervention; decision features, unique to the patient population in this case life-limited children; and process factors concerned with the wider context and health care environment.

Conclusions: Decision features, which include the evolving role of parents and young people as decision makers, and process factors such as the role of professionals and access to information for decision-making, can either enable or constrain families during the decision process and influence both the perceived choice and the degree of uncertainty they experience.

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- CHAPTER 1 -

INTRODUCTION, AIMS AND STRUCTURE

1.1 Background to the Study

This qualitative study examines decision-making for children and young people with life-limiting conditions for whom life-sustaining technologies can compensate for the partial failure or loss of a vital body function, and thus prolong life. The decision to administer technological support requires parents, professionals, and where appropriate children and young people to take into account complex issues regarding quality of life with respect to the likely progression and course of their condition (McHattie, 2005). As well as uncertainty regarding prognosis and the ethical and moral dilemmas families are confronted with when weighing up the long-term benefits and drawbacks, parents consistently report receiving inadequate and sometimes conflicting information and advice for decision-making (Spalding and McKeever, 1998; Craig and Scambler, 2006).

The evidence concerned with treatment decision-making for children with life-limiting conditions is sparse, and much of it anecdotal, drawing attention to the unique ethical and legal context within which these difficult decisions are made (Wright et al., 2009). It also highlights the differing opinions about the quality of life children with complex and often progressive conditions can achieve, a topic also discussed in the wider literature concerned with the use of life-sustaining technologies (Noyes, 2006a). Despite this, there continues to be a dearth of knowledge about the information and support needs of families for decision-making, and about how families and professionals share decisions of such complexity and uncertainty, and the consequences of doing so.

This exploratory study aims to shed light on the process Edwards and Elwyn (2006) refer to as the 'black box' of decision-making, and reflecting the applied nature of the research particular attention is paid to the information and support needs of

families. The study also focuses on the interaction between families and professionals for decision-making, acknowledging the important role professionals play in providing information, and the collaborative approach recommended for decision-making in the paediatric setting, promoted in both the National Framework for Children, Young People, and Maternity Services guidance for palliative care providers (Department of Health (DH), 2005), and the first ever Palliative Care Strategy for Children and Young People, *Better Care: Better Lives* (DH, 2008).

In acknowledging the central roles for parents, young people, and professionals in treatment decisions (Moore and Kirk, 2010), the study is underpinned by the framework of shared decision-making developed by Charles et al. (1999). This framework is grounded in the legally enshrined principle of informed consent that is central to health care decision-making (DH, 2001a; DH, 2001b), and recognises the political and cultural shift in Western societies from medical paternalism towards an informed and shared approach. Charles et al. (1999) also draw attention to the separate elements that make up the decision-making process, and the varying extent to which patients might participate in treatment decisions. The framework therefore enables decisions to be examined with reference to the key stages in the process, whilst still facilitating a flexible approach due to its acknowledgement of the complexity in treatment decision-making.

1.2 Introducing the Study

This study aims to offer a greater understanding of the decision-making processes involved in the administration of technological support for children and young people with life-limiting conditions, defined as “those for which there is no reasonable hope of cure and from which children or young people will die” (Association for Children’s Palliative Care (ACT), 2009). The diagnoses and prognoses for life-limited children and young people are wide-ranging, with some children living for only a few weeks or months, and others for years and even decades (Hynson et al., 2003). The need for technological support can occur at

different points in the illness trajectory, and whereas some children require the use of life-sustaining technologies at around the time they are born, others may not need support until they are teenagers or young adults (Wallis et al., 2011).

Therefore while it is likely that some decisions about technological support will be made during early infancy and others at the end of a child's life, this study is not about end of life decision making nor is it about the use of life-sustaining technologies for neonates. Similarly, this research is not about decisions that occur in a particular setting, even though some children with life-limiting conditions will be considered for technological support during a period of hospitalization and the decision process may be initiated within a Neo-Natal or Paediatric Intensive Care Unit (Wallis et al., 2011).

This research explicitly focuses on the use of technological support for children and young people who have been diagnosed with a life-limiting condition, whether or not the condition has an official label, and regardless of whether the need for technological support is life-threatening at the time a decision is made. It was important from the outset that the research, through exploring decisions for life-limited children and young people, was able to capture the diversity of experience among this small patient population, especially considering the growing acknowledgement of paediatric palliative care as a distinct speciality (Craft and Killen, 2007) and the recent policy drive to meet the needs of the increasing number of children diagnosed with life-limiting conditions in England (Fraser et al., 2011).

The focus on decision-making for life-limited children and young people also represents the applied nature of the research, which developed as a partnership between the Social Policy Research Unit at the University of York, and Martin House Children's Hospice, a key provider of paediatric palliative care in the Yorkshire and Humber region. The aims for the research arose primarily out of practice, and from the growing awareness among professionals who work with life-limited children and young people about the difficulties families experience when they are asked to make complex treatment decisions. Improving the information and support families

receive for decision-making was a key objective for the research at an early stage, and influenced the study design and aims as the research moved forward.

The collaborative nature of the research has been both a challenge and an advantage for the research, and the researcher has worked hard to ensure that the dual but at times conflicting goals for the research to generate new knowledge and provide practice based learning are met. This has in part been achieved by ensuring that the study aims and methodology have been informed by the existing literature as well as the insight provided by the collaborating partner about some of the complexities of decisions about technological support. While this knowledge was helpful in guiding the research design given the paucity of evidence in this area, it also meant that certain assumptions about the nature of decision-making among families of children with life-limiting conditions underpinned the early aims and objectives for the research.

The research process itself has also been influenced by the on-going collaboration between the researcher and the partner organisation, in particular the recruitment of families to the study and the overall process of data generation. The assistance of the collaborating partner in recruiting families to the study was invaluable, and the existing relationship between the researcher and Martin House enabled the research to achieve the desired sample. Martin House also offered additional resources for providing on-going support and training to the researcher, and for managing any potential distress suffered from participants as a result of taking part in the research. However, extra care was taken to ensure that the incentive to achieve the desired sample given the dual role of Martin House as partner and recruiter did not lead to bias within the sample.

1.3 Study Aims and Objectives

The central aims for the study are as follows:

- To explore how decisions about technological support are made by families
- To identify the key factors involved in decisions about technological support

- To explore the interaction between families and professionals during decisions about technological support.

Reflecting the applied nature of the study and in considering how the findings of the research might be used to inform future practice, two further objectives for the study were agreed at the outset.

- To identify the information and support needs of families for decision-making
- To explore the suitability of shared decision-making as a framework within which to understand treatment decisions for children and young people with life-limiting conditions.

Focusing on two types of technological support commonly considered for children and young people with life-limiting conditions, the decision processes explored for this study are:

- *whether or not to insert a gastrostomy tube, which enables a child to be fed when their condition has progressed to a stage where feeding and swallowing have become extremely difficult, and;*
- *whether or not to institute assisted ventilation when the child's breathing has become so constricted as to pose a risk.*

Given the paucity of evidence in this area, an exploratory approach was taken, and the study employed in-depth interviews with parents, and where appropriate young people to explore decisions they had made about gastrostomy insertion or ventilation, and in some cases both. During interviews, parents and young people were asked to think about which professionals were important in this process. These key professionals, with the family's permission, were invited to take part in an interview about their role in helping families with decisions about technological support. The family and professional data were analysed separately using the approach described by Spencer et al. (2003), which builds on the Framework

method of analysis developed by Ritchie and Spencer (1994) to meet the needs of applied and policy research.

Due to the depth and richness of data collected from families, and the nature of findings generated during analysis, this work reports on the findings from family data only. The framework presented is one derived from families' narratives of the decisions they have made and draws attention to unique features of decision-making for children with life-limiting conditions, as well as factors specific to decisions about technological support. The framework also incorporates the influence of professionals and the wider health care system in the decision process, and if the model is to inform future practice and policy it is essential that professionals involved in recommending treatments for life-limited children have an understanding of the decision-making process through the lens of families, especially if they are able to provide adequate information and support for decision-making. The decision to exclude the data collected from professionals in this research is underpinned by this goal.

1.4 Chapter Outline

Beginning with the existing evidence, Chapter Two provides an account of what is already known about the lives of children with life-limiting conditions and their families, drawing on the literature concerning the use of life-sustaining technologies and the wider literature concerned with children who have complex health care needs. An overview of the technologies investigated for the study – enteral tube feeding (more commonly referred to as gastrostomy), and mechanical ventilation, is then provided, drawing on the research exploring the impact on families as they take over the management of their child's technology in the home. Finally, the limited research concerned with the decision to administer life-sustaining technology for children with life-limiting conditions is discussed.

This leads on to the Chapter Three, which is concerned with the wider context of treatment decision-making, firstly providing an overview of the shift from medical

paternalism to informed and shared decision-making, and the impact of this shift on the practice of treatment decision-making in the UK. The second part of Chapter Three focuses on the ethical and legal context of paediatric decision-making, distinctive due to the child-parent-professional triad. Attention is then given to the practice of treatment decision-making in the paediatric setting, which is followed by the final section providing the rationale for underpinning the research by the model of shared decision-making.

Chapter Four presents the methodology employed for the research, with particular consideration to the research design, and to the ethical challenges experienced in the project and the nature of doing research in paediatric palliative care. As well as providing details of sampling and recruitment, Chapter Four provides an account of the data collection and analytical process. Finally, this chapter considers the rigour and credibility of the research, and provides an account of the factors considered in presenting the research findings.

Moving on to the findings, Chapter Five begins with an overview of the sample and details about the individual cases used for analysis before offering the reader a gentle introduction to the data, providing a number of contrasting case studies in order to illustrate the complexity of the decisions under analysis. Chapter Six presents a new framework through which to understand treatment decision-making for children and young people with life-limiting conditions, developed from the data and used to structure the chapters that follow.

Chapter Seven, Eight and Nine contain further details of the findings as they relate to the framework presented in chapter Six. Chapter Seven focuses on the decision factors used by families in weighing up the appropriateness of technological support for their child, chapter Eight on the features of decision-making unique to children with life-limiting conditions, and chapter Nine on the wider factors around the process of decision-making.

The final chapter draws out the main findings from the study, focusing on the ecological nature of treatment decision-making reflected in the new model, and

considering the potential consequences of the decision process for families as they continue to make on-going decisions for their child. This includes the potential conflict that can arise and how this relates to quality of life, which is viewed as a central factor in the decisions families make. The findings are then considered with reference to the existing framework of shared decision-making that underpins the research, with particular attention to whether the models of informed and shared decision-making reflect the way in which decisions for life-limited children and young people are made.

The chapter continues by setting out the strengths and limitations of the study, and considers how future research can further our understanding of treatment decision-making for children and young people with life-limiting conditions. The author then discusses the potential implications for policy and practice arising from the research, with specific reference to enabling families to make the right choices for their child before finally considering what the findings tell us about the model of informed choice that continues to drive forward healthcare policy in the UK.

- CHAPTER 2 -

TECHNOLOGICAL SUPPORT FOR CHILDREN AND YOUNG PEOPLE WITH LIFE-LIMITING CONDITIONS

This is the first of two chapters which aim to provide the reader with a background to the topic under investigation and an overview of the key points arising from the existing research and literature. This chapter begins with a summary of the approach taken to review the literature, which is followed by an account of the key themes concerning the use of life-sustaining technologies for children and young people, and the lives of children and young people with life-limiting conditions and their families. The chapter that follows this presents the literature concerned with healthcare decision-making, and making treatment decisions for children and young people.

2.1 Reviewing the Literature

Because of the existing work carried out to obtain funding for the research, the gap in knowledge about decision-making processes in the use of technological support had already been identified, and it was therefore deemed necessary to collect primary data in order to address this. The purpose of reviewing the literature again was to confirm the gap in knowledge identified during this phase, and to inform the current study. The review did not set out to identify and evaluate the existing evidence base. Instead a narrative review for each of the three key topics was carried out using a combination of key words identified from an initial review of the literature and from consulting with the supervision team (Timmins and McCabe, 2005; Cronin et al., 2008).

In reviewing the literature concerned with technological support for children and young people, particular attention was paid to the use of artificial nutrition and assisted ventilation, as the examples of life-sustaining technologies investigated for this study. Given the tendency in the literature to consider life-limited children

alongside other groups of ill children, the wider literature concerning children with complex health care needs was also included in the review. The literature search was conducted on MetaLib using ASSIA, Social Sciences Citation Index, CINAHL, PsycINFO, British Nursing Index, Medline and the Cochrane database (Timmins and McCabe, 2005). Only articles published in English were selected, and each search was limited to 1985 onwards. The titles were scanned initially to rule out clinical papers as these were not relevant to the topic under investigation. The remaining articles were then scanned by abstract, focusing on the aims and methods to identify the literature to be included for review.

To search the literature concerned with technological support, the keyword combinations “technological support” OR “technology dependent” OR “life-sustaining technology”; “gastrostomy” OR “enteral”; and “ventilation” OR “respiratory support” were each combined with AND “children” and then with AND “young people”. To review the literature concerned with life-limited children, the key word combinations “life-limiting” OR “life limiting” OR “life-threatening” OR “life threatening”; “palliative care”; and “complex health care needs” OR “complex healthcare needs” OR “complex care needs” OR “complex needs”, were each combined with AND “children” and then with AND “young people”. Truncating was applied to the terms child*, gastrostom*, ventilat* and life-sustaining technolog* to ensure all the literature was identified.

A different strategy was taken to review the literature concerned with decision-making due to the vast subject area and the different possible ways to approach the research with regards to decision-making theory. Initial discussions with Professor Patrician Sloper, the academic supervisor, were helpful in narrowing down the possible avenues for enquiry as she had been involved in a recent review of the literature for a separate project. The initial search strategy for their review using terms ‘choice’ and ‘decision making’ from 1985 onwards identified over 55,000 publications (Beresford and Sloper, 2008, p.55). Following consultation with Dr Dawn Dowding, an expert in the field of decision-making, their strategy for reviewing the literature was modified (Beresford and Sloper, 2008). This research

adopted a similar approach following consultation with the same expert, who was available for the current study in her capacity as Thesis Advisor.

The review of decision-making literature began with analysing the key texts concerned with decision-making theory, reports and policy documents regarding healthcare decision-making, and any existing literature reviews on the topic of healthcare decision-making. The main strands of literature were then identified and considered for use in the current research. Once the decision had been taken to focus on the conceptual and research literature concerned with shared and informed decision-making, a search of the literature using the same search parameters and review process described above was conducted using the key words “treatment decision-making”; “informed decision-making”; “healthcare decision-making”; and “shared decision-making” (the term ‘decision making’ was also applied for each key word using the Boolean operator OR). A separate search was performed using the same key words in combination with AND “pediatric*”; AND “paediatric*”; AND “child*”; AND “young pe*”; AND “adolescen*”.

2.2 The Use of Life-Sustaining Technologies

Advances in medical technology over the last half a century have led to the development of a range of medical devices to support individuals in hospital and at home whose condition or illness has caused one of their organs or bodily functions to deteriorate to the extent that it can no longer work effectively on its own. Examples of such devices include dialysis machines, gastrostomy tubes (a means of delivering artificial nutrition), and mechanical ventilators. Such devices are frequently referred to as life-sustaining technologies and are defined in the research literature as medical devices initiated “to compensate for the loss of a vital body function” (US Congress, 1987).

The advances in medical technology have developed alongside a societal shift towards home and community-based care for patients requiring long-term and complex packages of care, and a more recent drive to reduce the length of time

patients spend in hospital (Liley and Manthorpe, 2003). This drive towards care close to home has been welcomed by families and health care professionals who recognised long ago the positive outcomes associated with caring for children at home, and indeed was part of a wider campaign by patients with chronic conditions to be able to manage their symptoms while minimizing the effects on their daily routines (Thorne and Robinson, 1989; Heaton et al., 2003).

These developments have transformed the care of children and young people with complex health care needs, who in the past would spend prolonged and often unnecessary periods of time in hospital due to the extensive and often technical care required (Kirk, 1999). Consequently, parental participation in the care and treatment of ill children, of whom children with life-limiting conditions form a small sub-set (Craft and Killen, 2007), has become increasingly prevalent and a way of “reducing the adverse effects of hospitalization on seriously ill children” (Kirk, 1999, p.390). Combined, these developments have led to the emergence of a group of children and young people living at home “with continuing medical and nursing needs, some of who remain dependent on medical technology” (Kirk, 1999, p.390).

2.3 Technological Support for Children and Young People

The development of portable and clinically effective life-sustaining equipment means that an increasing number of children and young people with life-limiting conditions are not only surviving for longer, but are offered a better quality of life due both to the minimally invasive nature of technological support in the 21st century, and the potential for children to live a full and active life (Heaton et al., 2005). Children and young people requiring technological support are often referred to as ‘technology-dependent children’, a term applied over twenty years ago to define “a small sub-set of the disabled child population who rely on life-sustaining medical technology and typically require complex, hospital-level nursing care” (U.S. Congress, 1987, p.3).

Although it implies these children share common attributes, they are by no means a homogenous group, each having their own unique health care needs caused by a diversity of conditions, illnesses, or accidents, and accompanied by varying disabilities and impairments. Furthermore, where some children may require the occasional assistance of a mechanical ventilator, others require continuous support from a range of complex medical devices (Glendinning et al., 2001). While small, this group of children and young people is growing, partly because they are living longer due to their dependence on medical technologies, but also because the increased efficacy of such technology means that more children are able to benefit from the range of equipment and devices now available (Wallis et al., 2011).

Although the U.S. Congress definition is still applied today, much of the 'hospital-level' care referred to is now carried out by parents in the home, who are trained by health care professionals at the time technological support is initiated for their child (Heaton et al., 2003). As a result, an increasing number of parents whose children can now be considered for technological support take over many elements of care previously carried out by trained medical professionals (Glendinning et al., 2001). In response, we have witnessed an increase in the number of children's community nursing teams across the UK, who will often provide on-going support to the parents of children with complex health care needs (DH, 2008).

This study focuses on decision-making for two of the technologies often considered for children and young people with life-limiting conditions – gastrostomy (a form of enteral tube feeding), and mechanical home ventilation. They have been selected because of their contrasting features, and due to the complexities and uncertainties surrounding their appropriateness for children and young people with complex health care needs. Both technologies are managed predominantly by parents or other primary carers in the home, with and without support from health care professionals and others working in the community (Heaton et al., 2003).

2.4 The Use of Gastrostomies for Children and Young People

Medical and technological advances over the last thirty years mean that gastrostomy is now much safer and less invasive than it was when first administered to children, and the increasing number of children now fitted with a gastrostomy has resulted in the development of a range of portable and manageable devices for feeding in and away from the home (Gauderer, 2002). These changes mean that gastrostomy, as well as other forms of enteral tube feeding which were once considered as interventions to be managed in a hospital setting, are now predominately carried out in the home by patients and their families, usually with the support of primary care teams working in the community (Liley and Manthorpe, 2003).

2.4.1 Prevalence and indications

An increasing number of children with ongoing feeding difficulties are referred for a gastrostomy to prevent under-nourishment and help sustain normal growth and development (Gauderer, 2002). In fact, it is estimated that in the United Kingdom (UK) alone, there has been at least a 200 per cent increase in the number of children receiving gastrostomy tubes in the decade leading up to the 21st century (Herrington et al., 2001). In the United States (US), where the technology for performing percutaneous endoscopic gastrostomies (PEG) was developed in the late 1970's to increase the safety and effectiveness of gastrostomy feeding for children, it is estimated that around eleven thousand children now have PEGs (Gauderer, 2002).

The main indications for gastrostomy include vomiting, aspiration, reflux and difficulties swallowing, a condition known as dysphagia (Hament et al., 2001; Munro, 2003). Although there are multiple causes for these problems, some of which are extremely rare, they carry for all children a long-term prognosis of malnourishment, the consequences of which include growth failure and impaired neurological development (Royal College of Nursing (RCN), 2006). Children with a range of medical conditions are referred for a gastrostomy, including those with

degenerative conditions and life-limiting illnesses for whom gastrostomy tube feeding may be one of a number of medical technologies they come to depend on (Heaton et al., 2003).

Whether or not the difficulties swallowing and feeding relate to a gastrointestinal abnormality or are secondary to an underlying illness or disability, a gastrostomy is normally used to replace or supplement oral feeding, although it can also be utilised to provide fluids and medication (Sullivan et al., 2004). It is often recommended for children who need to be fed gradually, and many of these children will already be feeding through a naso-gastric tube (see Illustration 1). Unlike naso-gastric tube feeding, which is normally viewed as a short-term measure, a gastrostomy is usually recommended for children who require nutritional support for more than two or three months, and for some children and young people is a life-long intervention (Daveluy et al., 2005).

2.4.2 The Procedure and Equipment

For children, a gastrostomy is performed under general anaesthetic and involves creating an opening (stoma) into the stomach through which a feeding tube is inserted (see Illustration 2). Nutrients are then delivered via the tube directly into

Illustration 1

Illustration showing the position of a naso-gastric tube

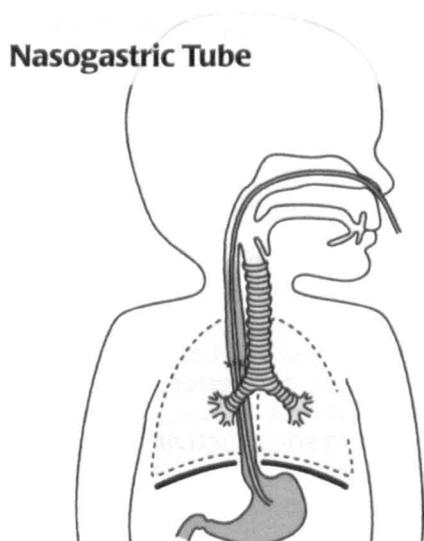
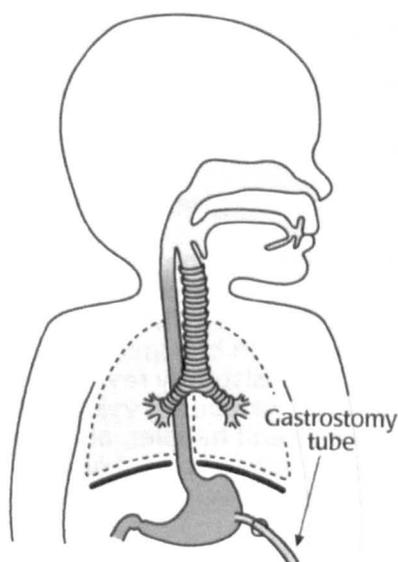


Illustration 2

Illustration showing the position of a gastrostomy tube



the stomach. Although there are several surgical procedures utilised to insert a gastrostomy tube, the majority are performed endoscopically, and the terms PEG, which refers to this less invasive procedure, and gastrostomy, are now more or less used synonymously (Gauderer, 2002).

In response to the growing demand for equipment that meets the diverse needs of children across the age range requiring a gastrostomy, there is now a range of feeding tubes and skin-level devices (often referred to as buttons) to choose from (Gauderer, 2002). Because the tube can be removed and the stoma closed, a gastrostomy can be both a permanent or temporary intervention (Great Ormond Street Hospital (GOSH), 2002).

Without post-surgical complications, children can return home in the days following surgery during which time parents will take over the management of their child's gastrostomy in the home. Children are either fed continuously over a number of hours, usually overnight, or at regular intervals throughout the day, a method known as bolus feeding. The method selected will depend on a number of factors unique to each child and their family. Feeds consist of special formulations tailored for each child in order to meet their nutritional requirements.

2.4.3 The benefits of a gastrostomy

There is a general consensus that enteral tube feeding for children with swallowing and feeding difficulties is beneficial, and as well as having a positive impact on physical growth and development, children are reported to be happier and more energetic (Craig et al., 2006). It is not surprising then to find that parents are generally satisfied with the outcomes for their child (Rogers, 2004). In fact, 86% of the 40 primary caregivers interviewed by Smith et al. (1999) "indicated that they would have started tube feeding earlier if they had known that it would have been so successful" (p.272). The growing studies exploring the parental experience of tube feeding consistently reveal parents to have few regrets, and report their child to be healthier and happier, and more able to participate in social activities due to increased energy levels (Smith et al., 1999).

Furthermore, many parents who prior to their child's gastrostomy invested huge amounts of time and energy feeding their child orally, report a number of positive changes to family life (Rollins, 2006). The challenge of feeding a child with swallowing difficulties can be particularly stressful, and parents often spend many hours each day to ensure their child receives adequate nutrition orally (Spalding and McKeever, 1998). In doing so, their child may experience frequent choking, stressful and prolonged feedings, and vomiting (Rogers, 2004). Once their child begins to feed effectively by gastrostomy, this distressing role is no longer, and some of the time previously spent on oral feeding can be set aside for more family oriented and social activities, and mealtimes are reported to be much easier for the whole family (Brotherton et al., 2007). In addition, parents are reported to be less anxious about whether or not their child is receiving adequate nutrition (Spalding and McKeever, 1998).

2.4.4 The drawbacks of a gastrostomy

The impact of gastrostomy feeding is not entirely positive, and Spalding and McKeever (1998) found mothers quickly "realized that G-tube feedings were 'mixed blessings'" (Spalding and McKeever, 1998, p.240). While the development of PEG, now the preferred practice for inserting a gastrostomy tube, is associated with a reduction in morbidity, there are still a number of major and minor complications associated with gastrostomy tube feeding in addition to the small risk of infection or bleeding with any surgical procedure (GOSH, 2002; Munro, 2003). For children with an existing respiratory problem or weakness, which is common among children and young people with a life-limiting condition, there is also an increased risk of respiratory, cardiac, and other problems due to the administration of a general anaesthetic (Morris, 1997).

Although the risk of major complications is small, with estimates ranging from 3 to 19% (George and Dokler, 2002), they can be life-threatening and may involve a hospital stay, and in a minority of cases additional surgery (Gauderer, 2002). Furthermore, gastrostomy feeding is associated with an increased risk of gastro-

oesophageal reflux, although the evidence to support this is not conclusive (George and Dokler, 2002). This risk is particularly worrying for parents involved in the decision for their child to have a gastrostomy because increased reflux can significantly affect the success of gastrostomy feeding, and for some children can involve further anti-reflux surgery to resolve it (Gauderer, 2002).

Amongst the minor complications, the stoma itself needs caring for regularly both during the weeks following surgery and over the longer term to prevent infection and soreness (Gauderer, 2002). Other minor complications include stomach cramps and swelling, sickness and diarrhoea, and accidental removal, leakage or blockage of the tube or skin-level device (GOSH, 2002). The incidence rate for minor complications is not conclusive, with estimates ranging from 13 to 23% (George and Dokler, 2002). However, while the rate appears low, it must be viewed with caution as all forty caregivers in Smith et al's. (1999) study reported at least one minor complication associated with tube feeding, although the definition of complication encompassed a wider range of factors than is normally the case.

2.5 The Use of Assisted Ventilation for Children and Young People

The technological advancements made over the last half a century have also enabled mechanical ventilation, which was once a complex procedure involving cumbersome machinery only suitable for hospital use, to become feasible for children and young people suffering from respiratory problems (Noyes et al., 1999). The development of non-invasive home ventilation has heralded a new era for children requiring ventilatory support, and although some children and young people require invasive surgery in the form of a tracheostomy, the advancements in this technology mean that home mechanical ventilation for children is now considered the norm, and the majority of children requiring assisted ventilation live at home under the care of a parent or guardian (Samuels and Boit, 2007).

2.5.1 Prevalence and Indications

Consequently, an increasing number of children with chronic or acute respiratory problems can be considered suitable for ventilatory support. Indeed the number of children relying on long-term ventilation, defined as a child or young person “who is medically stable and requires a mechanical aid for breathing either invasively by tracheostomy or by non-invasive mask interface for all or part of the 24 hour day” (Wallis et al., 2011, p.999), has increased over the last twenty years (Noyes, 2006a). It is also believed that as a result of technological advancements in recent years, the number of children and young people receiving ventilatory support will continue to grow, but at a more rapid pace due to the portability and increase safety of equipment now available (Noyes and Lewis, 2005).

Until recently, the exact numbers of ventilator-dependent children in the UK was not known. However a recent study by Wallis et al. (2011) has identified 933 children and young people living in the UK with long-term ventilation, 92% of whom lived at home at the time of data collection. The study was able to capture data about the majority of ventilator-dependent children in the UK, although admits that the real number is likely to be higher than this because some data were missing, and the study only identified children under the age of 17 years. However, the data provides evidence that the number of ventilator-dependent children has grown significantly from the 141 children identified in 1998. This study also confirms that many of the children considered for long-term ventilation have a life-limiting condition, although the exact number is not reported (Wallis et al., 2011).

The main indication for assisted ventilation is chronic and acute respiratory failure, and although a heterogeneous group, children tend to fall into three main categories – children with high cervical trauma following an accident or injury; children born with a congenital defect affecting the respiratory system; and children and young people with a neuromuscular disease (Noyes and Lewis, 2005). Of the latter group, young people with Duchenne muscular dystrophy, and increasingly children with spinal muscular atrophy (Ryan et al., 2007), both of which are

degenerative and life-limiting conditions, are among those considered for home mechanical ventilation, as their condition is associated with chronic and progressive respiratory problems (Wallis et al., 2011).

Some of the children and young people requiring assisted ventilation develop hypoventilation, the symptoms of which include poor sleep, depleted energy levels, failure to thrive, and recurrent airway infections (Nørregaard, 2002; Kinali et al., 2007). Other symptoms include a weak cough, retained airway secretions, and incompetent swallowing (Samuels and Boit, 2007). For many of these children, respiratory problems increase in severity over many years before they are considered suitable for support from a ventilator, and although early symptoms are often very subtle, there are a number of tests that can be carried out to determine the extent of respiratory problems (Samuels and Boit, 2007).

For some children though, hypoventilation is asymptomatic and the need for assisted ventilation is sometimes not detected until an acute episode of respiratory failure, which in most cases will lead to hospitalisation and for some children and young people, the initiation of assisted ventilation (Samuels and Boit, 2007).

2.5.2 *The procedure and equipment*

The purpose of ventilation is to provide oxygen to the body. The most common form of home ventilation is non-invasive positive pressure ventilation which is delivered through a small and portable machine (see Illustration 3 on page 35) via a face mask or a nasal mask. Some children and young people are not considered suitable for the non-invasive form of positive pressure ventilation, and will instead require a tracheostomy, a small opening in the windpipe which is held open by a tube for air to go in and out (see Illustration 4 on page 35) (Samuels and Boit, 2007). Depending on the symptoms and underlying cause, the needs of each child will vary with some children requiring the assistance of a ventilator at all times and others only requiring ventilation overnight.

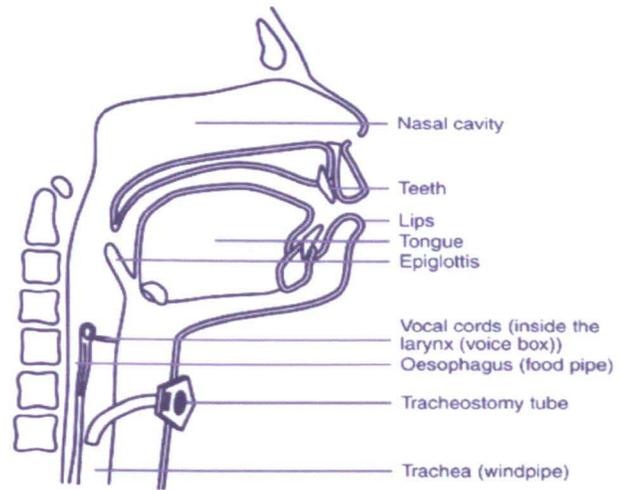
Illustration 3

Non-invasive Positive Pressure Ventilators
(adapted from Samuels and Boit, 2007)



Illustration 4

Illustration of tracheostomy
(adapted from GOSH, 2001)



The interface used will depend on a range of factors including age, likely compliance, and the underlying condition and symptoms (see Illustration 5). Correct sizing, position, and fit are also important, and patient preferences will be sought in order to enhance compliance (Samuels and Boit, 2007). There is little evidence about the effectiveness of different interfaces although Nørregaard (2002) suggests that for the paediatric population, “nasal masks seem to be the preferred type” (p.1334).

Illustration 5

Interfaces for paediatric ventilation
(Samuels and Boit, 2007)



Due to the personalisation of ventilatory support required for it to be administered effectively, the initiation of all types of ventilation normally requires hospital admission. With good planning though, children who will use non-invasive ventilation should be discharged from hospital in a few days, and for those requiring a tracheostomy around three weeks (Nørregaard, 2002). However, Nørregaard (2002) notes that meeting the needs of each child is complex, depending on a range of factors including interface,

compliance, resistance, pressure, frequency and potential leakage. In addition, some children require a humidifier where others do not. Therefore, prior to discharge “the child’s condition should be stable, the ventilation documented to be effective and satisfactory, and the family and attendants ready and motivated” (p.1335).

2.5.3 *The benefits of assisted ventilation*

There is a growing evidence base that the use of long-term ventilation for children is effective in managing acute and chronic respiratory failure (Hammer, 2000; Nørregaard, 2002; Samuels and Boit, 2007). In addition, Gilgoff and Gilgoff (2003) argue that “with proper follow-up care a high quality of life is a realistic expectation for ventilator-dependent children” (p.479), and this is borne out to some extent in the qualitative research focusing on the wider impact of living with a ventilator (Lindahl and Lindblad, 2011). Even for the young people in Noyes’ (2006a) study, whose ventilation was initiated towards the end of life, their health and quality of life was enhanced and they spoke about having more energy and generally feeling better (p.399).

For children with life-limiting conditions such as Duchenne muscular dystrophy, and increasingly spinal muscular atrophy, there is growing evidence that when administered at the right time it can be hugely beneficial, prolonging the life of young people by a number of years and enhancing their quality of life in many ways (Bach, 2008; Kinali et al., 2007; Simonds, 2007; Samuels and Boit, 2007). This has led to developments in the way conditions like Duchenne muscular dystrophy are managed, with regular monitoring now carried out to determine the optimum time at which to begin ventilation (Muscular Dystrophy Campaign, 2007).

Due to the positive outcomes associated with assisted ventilation for children, parents are reported to be satisfied with the outcomes for their child, who will experience better overall health and higher energy levels, enabling them to take part in activities not possible prior to the initiation of ventilation (Noyes et al., 1999; Noyes, 2006a). Moreover, the small studies exploring children’s views about their

ventilator-dependency reveal that they too are aware of the positive effects of assisted ventilation on their health, and other aspects of their lives (Noyes, 2006a; Earle et al., 2006). Both adults and children who depend on ventilation talk about the importance of the machinery in helping them live, and the incorporation of the technologies into their everyday lives (Lindahl et al., 2006; Earle et al., 2006).

2.5.4 The drawbacks of assisted ventilation

Although the benefits associated with having 'sufficient breath' can lead to a better quality of life, children and young people can also experience a range of negative psycho-social impacts relating to the level and complexity of care they require (Noyes, 2006a; Sarvey, 2008), and the loss of privacy and independence due to the presence of carers in their everyday lives (Sarvey, 2008). And whilst many children and young people are able to enjoy the sorts of activities and hobbies as their non-disabled peers, and in many ways 'normalise' their social experiences, there is also evidence that their equipment can become a barrier to taking part in childhood activities (Noyes, 2006a; Earle et al., 2006).

Furthermore, assisted ventilation is not without risks and complications. One of the major risks associated with managing ventilator-dependent children at home is accidental death, with recent estimates at around 4 to 5% (Schreiner et al., 1987; Nelson et al., 1996), and usually the result of an unobserved disconnection, machine failure, or airway accident (Noyes et al., 1999, p.442). The responsibility taken on by a primary care giver of a ventilator-dependent child, normally a parent, and in most cases mothers, is therefore immense, and for parents the risk of machine failure or accidental disconnection from the ventilator is overwhelming. In fact, the parents in Wang and Barnard's (2008) study described their child's ventilator as 'frightening but necessary', highlighting the vulnerability of children supported by mechanical ventilation.

Other complications of ventilation include aspiration, although the risk is minimal; air swallowing which can lead to abdominal distension; skin soreness and ulceration; eye irritation; facial flattening and deformity; and dryness of the upper

airways, a common complication solved by the introduction of humidification, and in fact many ventilators now come with their own humidifier (Nørregaard, 2002; Samuels and Boit, 2007; Markström, 2007). While non-invasive ventilation is preferred, some children and young people will continue to require a tracheostomy, and although complications such as skin and eye irritation are not a problem for these children, other complications are possible including loss of phonation, speech and swallowing difficulties, and infection of the tracheostomy site (Samuels and Boit, 2007).

Nevertheless, where long-term ventilation is monitored regularly, complications are generally minor, although there is a paucity of evidence to support this assertion (Nørregaard, 2002; Markström, 2007). Both Nørregaard (2002) and Samuels and Boit (2007) stress that for non-invasive ventilation to be successful, the correct sizing, fit and positioning of the interface is crucial. As well as minimizing the risk of complications such as skin soreness and facial flattening, and enhancing adherence by ensuring comfort and patient satisfaction, the correct interface also ensures minimal leakage and successful oxygenation. Samuels and Boit (2007) advise that sufficient planning should take place before a child begins ventilation so that time can be taken to select an appropriate interface which should be tailored to meet a child's psycho-social as well as health-related needs, as for some children the use of a full face mask can lead to feelings of isolation and detachment (Lindahl and Lindblad, 2011).

One of the key concerns regarding the institution of ventilation relates to the length of stay required in hospital, which for many children continues to take place in an intensive care setting due to the lack of appropriate facilities (Noyes, 2002; Ludvigsen and Morrison, 2003). Although the duration depends on whether or not ventilation is administered on an elective basis, or is instead the result of an acute episode, Noyes (2000b) points out that there are many additional and often unnecessary barriers preventing a timely and appropriate discharge from hospital, which for the children in Noyes' study adversely affected their experience of becoming ventilator-dependent.

The transition from hospital to home is therefore a highly emotive issue for parents, whose children they believe, are caught up in the tensions about where ventilator-dependent children should be managed and how quickly they can be discharged to the community (Noyes et al., 1999). There is some evidence of a shift in the way long-term ventilation is initiated, with some children and young people spending part of the initial period of ventilation initiation in the children's hospice setting as a step down between hospital and home, and in some cases at home with support from specialist children's community nursing teams or other healthcare professionals with expertise in the initiation of ventilator support (Nicholson, 2011). It is likely, that due to the increased safety and efficacy of ventilators, and the growing knowledge about the effective management of chronic respiratory problems, that this trend will continue.

2.6 *Caring for a Technology-Dependent Child*

Enabling children and young people requiring the use of technological support to live a full and active life is important to families, and Rabiee et al. (2005a) revealed a high degree of congruence between parents and children about the outcomes that mattered to them. Although generally in agreement, parents tended to focus on health, safety and communication first and foremost. In contrast, children and young people emphasised the importance of spending time with friends, of becoming more independent, of taking part in social activities, and of feeling 'normal' and doing the same sorts of activities as their non-disabled peers. Similar findings were identified by Kirk (2007), who found that "both the young people and parents displayed a determination to not let the condition or technology prevent them from living a full life" (p.3), with young people again emphasising the importance of living 'ordinary lives'.

However, the presence of technological support can have an impact on the extent to which this is realised, and Heaton et al. (2003), who explored "the rhythms and routines of caring for a technology-dependent child" (p.1), found that social schedules and activities were often organised around technical routines. Even

respite care and babysitters were scheduled for times when the care required for their child was minimal due to the difficulties in accessing competent carers. The research also found that the range of tasks falling under the realm of technical care was mainly carried out by parents, particularly mothers, and although they tended to be “structured around different natural, technological and social temporalities” (p.2), the time-demands frequently disrupted sleep and other activities, including children’s participation in school and social activities.

These findings mirror those of Kirk and Glendinning (2002) who collected interview data from parents of technology-dependent children and professionals involved in the home care of their child. They observed that “families’ lives often revolved around the technology and the routines it imposed” (Kirk and Glendinning, 2004, p.213), and as well as restricting the activities family members could participate in outside the home, they found that “the technology transformed the meaning of home, which were medicalized by the presence of equipment and the continual or frequent presence of home carers or professionals” (Kirk et al., 2005, p.459). They too found that for all families, “parents [are] responsible for the day-to-day management of their child’s condition and perform highly technical clinical procedures” (Kirk and Glendinning, 2002, p.625).

For children requiring artificial nutrition, parents consistently report a number of challenges associated with managing their child’s gastrostomy, particularly when there are complications such as increased vomiting, pain and gastroesophageal reflux (Brotherton et al., 2007). Spalding and McKeever (1998) found that even those parents who had felt prepared for the responsibility of caring for their child’s gastrostomy were overwhelmed by the immense changes it brought about in their everyday lives. The impact of such complications can also be exacerbated by the limited support parents feel they have as they begin to manage their child’s gastrostomy in the home, and the lack of adequate information they receive prior to their child’s gastrostomy, which can leave parents feeling isolated and overwhelmed (Brotherton et al., 2007).

Like the parents of children requiring a gastrostomy, parents of ventilator-dependent children can also find themselves overwhelmed by the immensity of complex and technical tasks associated with managing their child's equipment in the home (Noyes et al., 1999; Wang and Barnard, 2008). Sarvey (2008) interprets parents' reports of caring for a ventilator-dependent child as complex, with parents emphasising the importance of being able to care for their child at home but at the same time revealing the burden and isolation experienced as the primary carer of their child. Again, the nursing and social support families reported receiving was often inadequate with many families having unmet needs that prevented children and families from taking part in everyday activities (Noyes, 2006a).

The available research identifies the importance of training for parents as they take over the management of their child's new technology (Lindahl and Lindblad, 2011; Brotherton et al., 2007), which is argued to be essential considering the technical and complex tasks parents must carry out in order to minimize the risk of complications as their child adapts, and to ensure technological support is effective. Yet even with training, it is evident that this new role for parents can be overwhelming, particularly when follow-up support is inadequate or unavailable (Brotherton et al., 2007; Noyes et al., 1999). In fact Kirk (1999) highlights "the social, emotional and financial impact on families of providing intensive and complex nursing care for their child" (p.392), with social isolation, sleep deprivation, and feelings of stress and anxiety not uncommon. Wray and Wray (2004), as bereaved parents of a child with a degenerative condition, also highlight the effects of caring for a child with complex health care needs, noting that "perhaps the most difficult to cope with is the combination of disturbed sleep patterns and inadequate sleep" (p.204).

Although the immensity of the responsibility parents take on as carers of their child must be acknowledged, and the role they adopt supported, Reeves et al. (2006) stress that parent carers must be viewed as parents first and foremost, and not simply as carers of their child. This is supported by Beresford (1994a), who interviewed parents of a severely disabled child and found that parents often

rejected the label of carer. Nevertheless, carrying out complex nursing tasks on a loved one is very different to treating a patient, and the parents in Kirk et al.'s study (2005) describe undertaking such tasks as "the most distressing part of care-giving" (p.460). These parents described the tensions created by the dual role of parent and carer, which not only altered the meaning of parenting but for some led to "a sense of resentment at the way in which their nursing role could dominate their parenting experience" (p.460).

As Beresford (1994a) points out, "it cannot be disputed that parents of disabled children face a great deal of stress" (p.171), and as this assertion has become more widely accepted, research has turned to look at ways in which to alleviate the additional stress families of disabled children face. There is a danger though, that due to the emphasis on stress, health care professionals may view parents of disabled children as vulnerable and in need of help, when in actual fact many families actively seek out strategies to minimize the impact of their child's disability on their lives, drawing on the resources available to them (Sloper, 1998). Drawing on models of stress and coping to incorporate the heterogeneity of families' situations, research has begun to explore the different ways in which parents cope, and to identify beneficial coping resources and strategies families can utilise (Beresford, 1994a; Hodgkinson and Lester, 2002; Hildenbrand et al., 2011).

The way in which parents adapt to their new role can be seen among the parents taking part in Spalding and McKeever's (1998) study. Parents reported quickly developing the necessary skills to manage complications and problems associated with their child's new gastrostomy, and actively sought out solutions when problems occurred without necessarily having to rely on health care professionals. In fact, over time many of the mothers in this study "came to regard themselves as experts" (p.240), a finding reflected in the wider literature about parents who care for a child with complex health care needs (Wood et al., 2010; Kirk et al., 2005; Lindahl and Lindblad, 2011; Thorne and Robinson, 1989).

Nevertheless, the role adopted by parents in the home and the level of responsibility they assume can sometimes be overlooked by health care professionals and service providers who, as they accept this practice as standard, come to view parents as proficient and competent in caring for their child, despite the lack of comprehensive training which health care professionals previously expected to carry out the technical aspects of care have undergone (Reeves et al., 2006). There is also a concern that assumptions are made too easily about who will become the primary carer, and the level of care parents are able to take on over the long-term (Kirk, 2001; Sarvey, 2008).

2.7 The Role of Professionals in Supporting Families

While parents often come to view themselves as 'experts' in the care of their child (Kirk and Glendinning, 2002), the availability of appropriate support from professionals and service providers is identified as an essential resource for families of disabled children (Beresford, 1994a; Sloper, 1998; Kirk and Glendinning, 2004), and one that can both facilitate the process of coping by making available resources for families to draw upon, and reduce the potential of stressors to affect the lives of children and their families. In fact, the importance of the role of health care professionals and service providers is evident in all the research on technology-dependent children and other children with complex health care needs, and can greatly affect many aspects of the lives of children and their families (Emond and Eaton, 2004).

However, as well as being identified as a coping resource for families, one of the key stressors parents draw attention to is the inadequate provision of support, the fragmented nature of services, and the poor communication channels with health care professionals and service providers (Beresford, 1994a; Kirk and Glendinning, 2004; Heller and Solomon, 2005; Corlett and Twycross, 2006). Although there has been a growth in the number of community children's nursing teams across the UK, Kirk (1999) argues that "the emergence of this group of children in the community presents major challenges for health, social care and educational services" (p.390).

Not only do technology-dependent children require complex packages of care, which their parents are expected to deliver at home, but Noyes (2006b) points out that it is difficult for any one health care professional to have a complete overview of a child's package of care, particularly when there is an overlap between health, social care, and education.

Consequently, parents often taken on the role of managing their child's care package, which involves organising services and equipment, and navigating what can be a fragmented and inaccessible system in order to access care and support, a relentless task which they can find very time consuming and stressful (Kirk and Glendinning, 2002). Furthermore, the scope of technical care associated with their child's complex health care needs can make the task of sourcing respite and paid care in and away from the home particularly difficult because of the expertise required to manage their child's equipment competently (Heaton et al., 2003; Brotherton et al., 2007).

Health care professionals working with parents must therefore be aware of how the multiple role parents assume as carer, manager, decision-maker and advocate for their ill child "transforms their relationship with their child and their parenting identity" (Kirk et al., 2005, p.463). More over, Kirk (2001) argues, it is important that assumptions about the role of parents are not made by the professionals involved in supporting them, suggesting instead that a negotiation of the roles of both parents and professionals takes place, a recommendation also proposed by Shields et al. (2006). For Kirk (2001), this not only provides parents with a degree of choice and control about the level of responsibility they assume as carers of their child, but can also induce a more trusting and open relationship between parents and professionals.

Discussions and negotiations of this kind are much more likely to occur when there is an existing relationship of trust and continuity between the family and care team, with continuity of care and family-professional collaboration highlighted in several studies as facilitators to achieving this (Beresford, 1994a; Heller and Solomon, 2005;

Pontin and Lewis, 2009). Heller and Solomon (2005) found that staff continuity “facilitated communication and sharing of expertise about their child’s condition” (p.340). They also revealed that “in the absence of continuing and caring relationships with staff, parents expressed frustration, hypervigilance, and mistrust about the quality of care that their child was receiving” (p.343).

For parents of children with ongoing health care needs, the relationship they have with the health care system is on-going, and the trust they place in health care professionals is an evolving process rather than a static feature (Thorne and Robinson, 1988a/1988b/1989; Kirschbaum and Knafelz, 1996). Thorne and Robinson (1989), using the findings from a qualitative study with chronically ill patients and their families, developed a model portraying the fluidity of health care relationships, which incorporates three key stages – naïve trust, disenchantment and guarded alliance. This process involves families moving from a position of what Thorne and Robinson (1988b) refer to as ‘blind faith’, towards a trust constructed “on an informed rather than a naïve perspective of the skills and limitations of professional care” (p.783-4). During this transition, families experience the loss of initial trust and therefore a period of disruption to their relationships with professionals. Their position on the trust continuum is also affected by their level of competence in managing their child’s condition (Thorne and Robinson, 1989).

Thus, the process of negotiating the respective roles of parents and professionals is fraught with difficulties, and Kirk and Glendinning’s study (2004) found that parent-professional relationships worked best when professionals openly acknowledged parents expertise and knowledge in caring for their child, and also the emotional costs involved. It is also argued that despite the blurring of parent and professional boundaries, and the growing expertise of parents in ‘nursing’ their ill child, nurses, and in particular children’s community nursing teams continue to have a key role to play in supporting families of technology-dependent children, both during the child’s stay in hospital and following their return home (Kirk and Glendinning, 2004).

However, due to the transformation of the parent-professional relationship caused by the evolving role of parents as they become experts in the care and treatment of their child, Kirk and Glendinning (2002, 2004) argue that the role of community-based nurses needs redefining if they are to effectively meet the needs of technology-dependent children and their families. As a result, rather than traditional nursing support, parents and professionals agreed that emotional support, instrumental and practical help, and information were the most needed and valued, although “nurses considered that part of their role was to ensure that parents performed clinical procedures safely” (Kirk and Glendinning, 2002, p.629).

2.8 Making Decisions about Technological Support

Both the role assumed by parents as primary carer of their child, and the evolving relationship dynamics between parents and health care professionals have the potential to influence how decisions are made for children, particularly given the finding in Kirk and Glendinning (2002) that in many cases “parents rather than professionals are the experts in the child’s care” (p.628). As well as being responsible for making the decision for their child to become dependent on technological support, parents also become responsible for the outcomes of the decision as they take over the management of their child’s technology in the home. Any uncertainty or regret associated with this decision may well be exacerbated by the dual role they assume as parent carers, particularly when there are complications and problems they have to resolve.

Parents can experience emotional factors in the decision for their child to use technological support as well. For parents of children requiring a gastrostomy, the thought of moving from oral to tube feeding can be associated with ‘loss’, the consequences of which parents perceive to include losing a source of pleasure, socialisation with others, family mealtimes, and child-parent interaction (Rollins, 2006). When a child is considered for tube feeding, parents are often unsure about the impact of such consequences, and Guerriere et al. (2003) found some parents believed oral feeding would cease altogether once tube feeding began. This

perception is worrying given that “most children can safely enjoy oral feedings or tastes in conjunction with tube feedings” (Rogers, 2004), p.31).

As well as a sense of ‘loss’, parents often associate the decision to go ahead with a gastrostomy for their child as ‘giving in’, and Rouse et al. (2002), whose study focused on children with complex health needs, point out that “feeding routines are one of the last ‘normal’ parent-child interactions for these families, and changing them can be problematic because of strong emotional and attitudinal ties” (p.123). For parents a gastrostomy “signified not only that they had failed in their role as nurturers but that their children’s disabilities were severe” (Spalding and McKeever, 1998, p.239). This was reflected by Rollins (2006) who found that the experience of caring for a child with feeding difficulties can “threaten a mother’s identity as a ‘good mother’” (p.19), and points out that “mothers often express a feeling of culpability for their child’s poor growth and the need for surgery as a failure” (p.31).

Rogers (2004) highlights other concerns parents express about dependency, safety and the invasive nature of a gastrostomy, and for the parents in Spalding and McKeever’s (1998) study, gastrostomy “was considered another invasive procedure with stigmatizing properties that they did not want their children to undergo” (p.239). For decisions involving older children, additional concerns around independence, self-image and social interaction are likely to arise (Ali et al., 2005), although there is no published research exploring the views of young people with a gastrostomy.

Research concerned with decision processes in the use of mechanical ventilation for children and young people is sparse, and much of it offers, from an ethical and medical perspective, the decisions as they relate to the critically ill or severely disabled neonate or child, therefore viewing the decision solely as an end of life one (Street et al., 2000; Edwards, 2002; Vrakking et al., 2005; Fortune, 2006; April and Parker, 2007). Although decisions about technological support for children and young people with life-limiting conditions are sometimes made at the end of life, the circumstances under which these decisions are made is very different because

they are as much about enhancing quality of life as they are about extending life, and are often underpinned by the principles of palliative care that guide the management of a child's condition (Fraser et al., 2010).

2.9 Making Decisions for Children with Life-Limiting Conditions

For parents of children with a life-limiting condition, the invariable uncertainty regarding the prognosis for their child creates a further dilemma in the decision process, bringing to the fore the issue of quality versus length of life (Wright et al., 2009). Therefore, as well as weighing up the benefits and risks in order to make an informed decision, parents and clinicians can find themselves involved in moral and ethical discussions about the appropriateness of technological support. Combined with this prognostic uncertainty, making a decision regarding technological support is a complex process, and for young people with degenerative conditions these decisions often come at a time when the disease has progressed significantly and palliative care planning is very much on the minds of those involved (Wright et al., 2009).

Moreover, while children and young people with life-limiting conditions form only a small subset of the children requiring technological support, they can often require the assistance of at least one device due to the progressive nature of their condition (Heaton et al., 2005). Some children may not require the assistance of life-sustaining technology until later on in their lives, while others may depend on technological support for most of their lives. In fact, parenting a child with a degenerative and life-limiting condition is unique because the tasks of caring can become more complex over time as "the symptoms gradually become more numerous, more severe and increasingly unpleasant and distressing" (Wray and Wray, 2004, p.201). In addition, children with life-limiting conditions follow their own distinctive illness trajectory, and their health care needs will continually change throughout their lives (Aldridge, 2007; Craft and Killen, 2007).

Childhood life-limiting conditions are extremely distressing for families because they conflict with all that we associate with childhood (Hynson et al., 2003; Aldridge, 2007). In fact, as Aldridge (2007) points out “nothing, absolutely nothing, prepares us for the compromise of our child’s life” (p.2). Inevitably, when a child is diagnosed with a life-limiting condition for which there is no known cure, their life, and the lives of those closest to them are changed forever as they embark together on an uncertain and painful journey over which they have little control (Steele, 2002; Hynson et al., 2003). These children and young people not only live with conditions for which there is no known cure, but they also have a shortened life-expectancy, with some children living only a short time, and others for years and sometimes decades (Sourkes et al., 2005).

Parents cope with their child’s illness in different ways and to varying degrees, and the sorts of coping resources and mechanisms families adopt can influence the way in which they are able to make, and participate in decisions regarding the treatment and care of their child. Some parents will continue to seek for a cure and push for aggressive treatments throughout their child’s life, even at a time when these treatments offer little or no benefit (Gillis, 2008). Others may accept that their child at this stage requires only palliative care and symptom relief, but will find it immensely painful to do so (Aldridge, 2007). What is clear from the parents in Steele’s (2002) study, is that “families need guidance and support from professionals in a collaborative relationship that extends beyond the boundaries of hospitals and institutions” (p.433).

The complexity of decisions regarding gastrostomy and assisted ventilation for children with life-limiting conditions whose condition will continue to deteriorate despite the benefits associated with technological support, are exacerbated by growing concerns within the medical community that because of their increased simplicity and safety in the form of PEG and non-invasive ventilation, they are becoming over-utilized for children, particularly at the end-of-life (Gauderer, 2002; McHattie, 2005; Noyes, 2006b; Ryan et al., 2007). This concern, combined with the uncertainty about the appropriateness of technological support for children

towards the end of life, can lead to both intra-personal and inter-personal conflict in the decision-making process, especially when individual values about quality of life amongst those involved in the decision diverge (Wright et al., 2009).

Furthermore, as the lives of children and young people with complex health care needs are extended, the decision-making process is surrounded by further uncertainty regarding the new and unexpected complications associated with living much longer lives (Samuels and Boit, 2007). These uncertainties, whether the result of new technologies and therapies which enhance longevity, or due to the sometimes rare or undiagnosed conditions associated only with childhood, add a complex dimension to the treatment and management of children with life-limiting and progressive conditions, and about the appropriateness of technological support, both prior to initiation and afterwards as a child's condition continues to deteriorate.

These concerns about the appropriateness of life-sustaining technologies reflect a wider debate about what is in the best interests of children and young people with complex health care needs and severe disabilities, many of whom will have a life-limiting or life-threatening condition (Noyes, 2006b; Wilkinson, 2006). Although there is a small evidence base that children are living longer and with a higher quality of life than was possible in the past, there is a need for greater research in this area, particularly given that there is some evidence suggesting "differences in perceptions between doctors, parents and affected children, with children valuing their health and quality of life more highly" (Noyes, 2006a, p.394).

The findings from these studies are important because they begin to challenge some of the views, often seen in the mass media, about the use of life-sustaining technology for children (Seymour, 1999; Noyes, 2006a). High profile cases are often contextualised in the media with regards to the on-going polarized debate about the use of life-sustaining technologies. On the one side, it is argued that the existence of technology-dependent children should be seen a 'failure of medical science' and a drain on finite resources. On the other, it is believed that all "children

have a right to life, and a right to resources and nursing services" (Noyes, 2006a, p.393), and that their health-related quality of life should not be judged in such simplistic terms (Stanley, 2000). In addition, the use of medical technology at the end-of-life is often argued to be 'unnatural' and 'inhumane', which Seymour (1999) notes, for some, poses a "threat to an idealized 'natural' death" (p.691).

This debate is not, however, restricted to the media. It also takes place when children and young people are considered for support from life-sustaining technologies, during which a child's quality of life is a key factor (Wright et al., 2009). In cases where two or more of those involved in the decision (in most cases the child, the parent/s, and the clinician) may disagree about the best course of action, it is matters regarding the child's quality of life that will often be the cause of such conflict (Stanley, 2000). Wright et al. (2009) point out that while conflicts around quality of life "are usually worked out in a considered and constructive way between the various parties involved" (p.238), there will be instances where it cannot be resolved. It is in these cases that a third party may become involved to facilitate a solution, or that the courts may be called upon "to make an objective, independent decision" (p.246).

Wright et al. (2009) draw attention to the fact that "those situations that reach the media and courts are not representative of everyday practice" (p.246).

Nevertheless, Guerriere et al. (2003), who explored decisional conflict among families whose child required a gastrostomy, found that half of the fifty mothers in their study felt pressured by health professionals to either refuse or agree to a gastrostomy for their child. Furthermore, around a quarter felt they were not supported during the decision-making process (p.475). Thorne et al. (1997) sum it up succinctly, describing the decision as "highly context-dependent, replete with multiple meanings, and typically complicated by avoidable miscommunications and misunderstandings between parents and health care professionals" (p.98).

It is therefore imperative that families are enabled to make a fully informed decision regarding the administration of technological support. Yet Noyes et al.

(1999) remind us that “understanding and meeting psychosocial issues facing ventilator-dependent children and their families has lagged behind advances in technology and medical expertise” (p.449), and quite often the information and support required by families during the decision-making process is fragmented and insufficient. Herrington et al. (2001), reviewing the literature on gastrostomy for children, also found “the support given to the family prior to, and after the operation is inadequate” (p.375), and Todd et al. (2005) raised concerns that “such decision-making regularly occurs in an environment of major emotional stress and with inadequate information about the device, method of placement, associated risks or the likely clinical course” (p.188).

This is a worry, because providing parents with appropriate information and support during and beyond the decision-making process is argued to be positively correlated with ‘familial psychological and emotional wellbeing’ (Rouse et al., 2002, p.123). Moreover, the communication of information is an essential component of the decision-making process for families, as it can help parents address some of the uncertainties they experience, and to ensure parents, as primary care givers for their child, are adequately prepared to take over the management of their child’s equipment in the home. However, despite the centrality of communication in the process of information and support giving, Levetown (2008) note it “is a critical, but generally neglected, component of pediatric and pediatric subspecialty practice” (p.1441).

2.10 Chapter Summary

Children and young people with life-limiting conditions form a small sub-set of the larger number of children who require the use of technological support, and face unique challenges due to the prognostic uncertainty and life-shortening nature of their condition. The literature in this area is expanding, and certainly during the last twenty years there has been a growing body of research concerning children and young people with life-limiting and life-threatening conditions, and those with complex healthcare needs. Before drawing some relevant conclusions from the

literature and evidence introduced in this chapter, the key points are summarised below.

- Caring for a child with complex health care needs is a relentless and challenging undertaking accompanied by many adverse effects and consequences. Parents are often overwhelmed by the immensity of tasks involved in managing their child's equipment in the home, and experience a great deal of stress which go beyond the nature of caring for a child with complex health care needs, and the impacts of which include poor health-related quality of life and lower levels of well-being.
- The psycho-social and emotional stress of parenting a child with complex health care needs, and the coping resources available to families, can affect the ways in which they are able to participate in decisions about technological support. Furthermore, the dual roles parents assume as primary carer of their ill child are often at odds to one another, causing tension for parents as they take over the management of complex nursing tasks in the home.
- As the knowledge and expertise of parents increase, their expectations of health care and other professionals involved in caring for their child change, as does their need for information and support. The traditional boundaries associated with parent-professional relationships are blurred, influencing the roles they each play in the child's care and treatment, and also in decision-making.
- Only a minority of the children and young people referred to as technology dependent have a life-limiting condition, and the technological support required by these children occurs under very distinct circumstances, and often at a time when their prognosis has worsened and yet their need for technological support is increasing. The decision families and health care professionals make about technological support is therefore clouded by the uncertainty surrounding a child's prognosis, and the unknown consequences of extending the lives of children and young people than was previously possible.

- Quality of life is an important factor for parents, young people, and health care professionals when considering the benefits of technological support. However, perceptions vary as to what 'quality' means, and it is believed that the views and wishes of children and young people, whose quality of life will undoubtedly change as a result of administering technological support, are often overlooked or secondary to the more tangible outcomes associated with a given treatment.
- It is believed that there are conflicting opinions within and between families, and between health care professional groups and individuals, about the appropriateness of technological support for children and young people with complex health care needs, reflecting a wider societal debate about the appropriateness and the costs involved of prolonging the life of children and young people, and their supposed 'quality' of that life. This, it is argued, can lead to conflict in the decision-making process, and parents can feel under pressure to make decisions based on professional recommendations.
- Families report receiving inappropriate information and support, both during the decision-making process and as they begin to manage their child's medical equipment in the home and community. This reflects wider concerns about the lack of appropriate support for families of children with complex health care needs.

The literature presented here suggests that the recommendation health care professionals make to offer technological support for a child or young person with a life-limiting condition, and the decision parents make with or for their child about whether or not to proceed, is a complex decision surrounded by uncertainty about their child's prognosis, their quality of life, and their wishes and needs. The literature also implies that discussions about quality of life are often at the crux of such decisions, and that disagreements about the meaning of 'quality' in these difficult circumstances can arise, sometimes between families and health care professionals, and sometimes within families or health care teams.

However the evidence base is small and the review identified a skew towards primary research that either considers the medical efficacy of a particular intervention, or focuses on the experiential accounts of families as they manage their child's condition in the home. Although some of the experiential research identified key points for decision-making, there was no research that focused on the process of decision-making itself. Some of this research is also quite old, and given the pace of medical advancements in recent years, combined with changes in the way healthcare is delivered in the UK, they are likely to be out of date. In addition, some of the studies are poorly designed and in many cases involve small convenience samples, partly because of the low numbers of life-limited children and young people, and also due to the difficulties in conducting research in this area (Stevens et al., 2010).

The second strand of literature identified from the search includes review articles, opinion pieces and descriptive papers that tend to focus on the ethical and moral challenges of making decisions for children and young people with complex health care needs. Where explicitly relevant to the topic under investigation, this secondary evidence was included in the review as it offers an important insight into the nature of decision-making around the use of technological support. However, it is important to note that some of the articles are presented from the perspective of the authors, and for healthcare professionals, from their experience in working with life-limited children and their families.

In considering the overall quality of the literature, and examining the key points identified from the review, it is evident that there is a gap in knowledge about how families make decisions about technological support for children with life-limiting conditions and children with complex health care needs more generally. The following chapter therefore focuses on the theories and models of informed and shared decision-making in order to learn more about the process of making treatment decisions, and the way in which parents, children and health care professionals interact during this process.

- CHAPTER 3 -

EXPLORING THE PROCESS OF TREATMENT DECISION-MAKING

Chapter Two drew attention to the increasing use of technological support for children with complex healthcare needs, of which life-limited children and young people form a small sub-group. The chapter also discusses the nature of making decisions about technological support, and the unique characteristics of life-limited children that might influence the process of decision-making. This second literature chapter aims to provide the reader with an account of how treatment decisions are made for children and young people, and the following section provides further details about the approach taken to review the decision-making literature, and the topics introduced in this chapter.

3.1 Approaching the Decision-Making Literature

The literature concerned with decision-making is vast and multi-disciplinary, with relevant material about the way individuals make decisions evident in the fields of philosophy, psychology, economics, sociology, health, and social policy (Koehler and Harvey, 2007). Therefore determining the relevance of different approaches within the literature to the process of treatment decision-making was a mammoth undertaking, and one that was informed by the key points covered in Chapter Two about the lives of children and young people with life-limiting conditions and their families, and the sorts of decisions they will make about the care and treatment for their child's related symptoms and difficulties.

The limited available knowledge concerned with the process of decision-making around complex medical interventions and the exploratory nature of this study also guided the literature review to ensure that the approach underpinning the research was not based on certain assumptions about the processes under exploration. Therefore, while the psychological theories and concepts of decision-making were particularly useful in considering the types of strategies and heuristics individuals

use in processing information for decision-making, and also introduced key concepts such as the cognitive continuum, anticipated regret, cognitive dissonance, and decision uncertainty (Beresford and Sloper, 2008), no single approach considered very complex treatment decisions, and much of the work focuses on the individual as opposed to the wider process of decision-making around the individual as they weigh up the choices presented to them.

The philosophical literature was valuable in provoking a thoughtful consideration of the assumptions that underpin the concept of informed consent, a central component in the process of treatment decision-making (Buchanan and Brock, 1990; Friedman Ross, 1998; Jonsen et al., 2006). However, again much of the literature, while useful, focuses on the autonomous individual, and the principles underpinning the process of making an informed choice. Nevertheless, this overlapped with the literature concerned with the political and ideological shift across many Western societies towards models of treatment decision-making that recognise the principles of autonomy and free will, culminating in the models of informed and shared decision-making (Charles et al., 1999). While much of this literature is about 'participation', a small sub-set is focused on the actual 'black-box' of decision-making, and what happens within it when the parties involved are the patient and recommending physician (Edwards and Elwyn, 2009).

This chapter will therefore position the process of making decisions for children and young people within this paradigm, one that draws attention to both the patient and referring professional, and also the institutional and legal system within which decisions are made. Given the applied nature of the study and the objective to identify the information and support needs of families, the focus on considering the whole process and not simply the way in which families weigh up the use of technological support is essential. However, in choosing to explore the overall process, it is important that this study does not become about participation in decision-making, because while it is acknowledged as a central element, it is still only one aspect of a much more complex and ecological process.

This chapter therefore presents the literature concerned with the legal and ethical institution within which treatment decision-making occurs, focusing on the shift from medical paternalism to shared and informed decision-making, and the research concerned with the interaction of patients and professionals in making decisions. This provides a backdrop to the similar transformation in the way treatment decisions are made for children and young people, which will be introduced, drawing attention to the relevant legislation and policy initiatives influencing this development. The remainder of the chapter will focus on treatment decision-making for children, acknowledging the triadic and unique legal context in paediatrics, before providing a rationale for underpinning the research by the model of shared decision-making, which is used as a guiding framework for the current study.

3.2 From Medical Paternalism to Informed Decision-Making

The concepts of shared and informed decision-making, which encourage patients, and in the case of children their parents, to participate in and take more responsibility for decisions affecting their treatment and care, have now permeated both the primary and acute health care sectors, and are actively promoted within the NHS (DH, 2001c). In fact, the rhetoric of patient participation has infiltrated every area of policy and practice in health and social care in the UK (Thompson, 2007). This transformation, commonly referred to as the shift from medical paternalism, provides an important context to the reality of treatment decision-making in the 21st century because of the dominance of paternalism, exemplified by the active-passive doctor-patient relationship, for more than two hundred years (Kaba and Sooriakumaran, 2007).

3.2.1 The rise and fall of medical paternalism

The rise of the biomedical model of illness during the 19th century, which was built upon the increasing volume of medical knowledge and expertise during that period, “focused not on the symptom, but rather on the accurate diagnosis of a pathological lesion inside the body” (Kaba and Sooriakumaran, 2007, p.59). Moving

away from viewing the patient as an individual, and instead emphasising the importance of biology, physiology, and medical expertise in the process of diagnosis and treatment, the biomedical model began to influence the practice of medicine and enhance the status of physicians, whose role it became not to simply treat the patient's symptoms, but to examine the patient's body, applying their knowledge and expertise in order to formulate a medical diagnosis (Kaba and Sooriakumaran, 2007).

This process altered the relationship between the physician and patient, which during the 1700s was dominated by the patient, and had in fact shifted along the patient – physician control continuum for many centuries, influenced by the prevailing model of illness at the time (Kaba and Sooriakumaran, 2007). As the biomedical model continued to shape medical practice during the 19th century, the physician began to dominate the doctor-patient encounter, further enhancing their status in the relationship. This new model of the doctor-patient relationship, referred to as 'medical paternalism', and recognising in its label and conceptualisation the inherent power and information imbalance therein, portrays the type of relationship between a dominant (active) doctor and a passive patient, who, due to their medical ignorance, is entirely dependent on the knowledge and expertise now associated with the medical professional.

Sometimes referred to as the parental or priestly model of physician-patient interaction, Emanuel and Emanuel (1992) argue that in the paternalistic relationship, the physician's sole aim is to promote health and well-being. By using the knowledge and expertise gained from professional training, the physician can therefore "discern what is in the patient's best interest with limited patient participation" (p.2221). Consequently, patient autonomy is disregarded in the process of decision-making, and the patient's values and preferences are assumed to match those of the physician, which is to "ensure [they] receive the interventions that best promote their health and well-being" (p.2221).

In its most extreme form, this model represents a process in which “the physician authoritatively informs the patient when the intervention will be initiated” (Emanuel and Emanuel, 1992, p.2221), and in which “the patient will be thankful for decisions made by the physician even if he or she would not agree to them at the time” (p.2221). However, in reality paternalism comes in many forms, and Emanuel and Emanuel (1992) remind us that, while more commonly depicted as the extreme form of activity-passivity illustrated above, in many decision-making scenarios described as paternalistic the physician may well provide the patient with information about a proposed intervention or treatment, and also seek their consent, although it may not be informed.

In fact, the point at which paternalism ends and another type of interaction begins, remains contested, with a number of authors preferring instead a continuum with physician control, or medical paternalism, at one end, and patient control, or informed choice, at the other (Degner, 1998; Guadagnoli and Ward, 1998; Thompson, 2007). While these authors may employ different concepts to portray the patient-physician decision-making continuum – control (Degner, 1998); power (Thompson, 2007); and participation (Guadagnoli and Ward, 1998) – they all argue that such an approach offers a more realistic perspective of the diversity in decision-making, as opposed to the ‘ideal type’ associated with a conceptual model.

During the second half of the 21st century, the idea of physician control and dominance came under heavy criticism by both the patient and medical communities, the former calling for patient autonomy and choice in the process of treatment and care, partly in response to growing evidence about the quality of medical care (Emanuel and Emanuel, 1992; Charles et al., 1999), and the latter beginning to recognise that a different approach would be required to effectively meet the needs of the growing number of patients living with an illness for which there was no single best treatment (Charles et al., 1999).

The models being promoted to address these challenges, the most widely discussed being informed choice and shared decision-making “were developed largely in

reaction to the paternalistic model and to compensate for alleged flaws in the latter approach” (Charles et al., 1999, p.654). Furthermore, they are underpinned by the process of informed consent, which in the UK and other Western societies is “now ethically and legally ensconced as a patient right” (Charles et al., 1997, p.681).

3.2.2 *The model of informed choice*

The model of informed choice has at its core the conception of patient choice, and is underpinned by the principle of patient autonomy. It acknowledges that for conditions involving trade-offs between treatment options, “the patient rather than the physician would have to live with the consequences of these trade-offs” (Charles et al., 1999, p.653). Otherwise referred to as the informative, scientific, or consumer model (Emanuel and Emanuel, 1992), the informed model is positioned at the opposite end of the patient-physician decision-making continuum to paternalism, because ‘decisional control’ now belongs to the patient rather than the physician (Thompson, 2007). However, while passivity belongs to the physician in terms of decisional control, the physician’s role is still central to the decision-making process as the primary goal shifts to one of “providing the patient with the means to exercise control” (Emanuel and Emanuel, 1992, p.2221).

For Thompson (2007), this requires “a narrowing of the information / competence gap between professional and patient, with some surrendering of power by the professional” (Thompson, 2007, p.1299). The physician’s role therefore, is reduced to one of information provider, offering the patient unbiased and clear information about their diagnosis, the proposed treatment or intervention, and the possible risks, benefits and outcomes. The patient, now equipped with the technical and medical knowledge of the trained professional, is expected to evaluate the different types of trade-offs for each possible treatment, including the option of no treatment, to make an informed decision, free from persuasion or coercion (Emanuel and Emanuel, 1992).

Bekker et al., (1999), who define an informed choice as “one where a reasoned choice is made by a reasonable individual, using relevant information about the

advantages and disadvantages of all the possible courses of action, in accord with the individual's belief" (p.1), argue that while such an approach incorporates patient values within the decision-making process, it makes an assumption that patients not only have a clear and fixed idea of their values and preferences, but are able to interpret the meaning of these for the decision in hand. As the role of the physician is limited to one of information provider, the patient ultimately carries responsibility for the final decision, having made an informed and voluntary choice based on the information provided.

This definition of informed choice carries an implicit assumption, again in contrast to the model of paternalism which suggests the patient has no capacity for decision-making as a result of their condition and medical ignorance, that the patient is in fact a rational actor with the capacity to process what can be complex medical information about the benefits and risks of a proposed intervention, and combine this information with their own values in order to make an informed decision. This assumption reflects the broader work of decision-making theorists, particularly those sharing the normative approach to decision-making which, supporting the notion that human beings use logic and reason in the process of decision-making, offer us "rules to follow or conform to that supposedly make our thought rational" (Over, 2004, p.3).

A further supposition in this model, also underpinned by the normative approach to decision-making, is that the physician, as a logical and rational human-being albeit one holding extensive medical knowledge, is able to become the effective provider of information for decision-making. Yet the ability to provide objective and clear information to patients requires excellent communication skills and knowledge of how individuals process information for decision-making to avoid coercion and bias, particularly when there is uncertainty regarding the benefits and risks of the proposed intervention (Bauchner, 2001). Furthermore, the idea that the physician enables the patient to assume control by giving away power and providing information is important because it implies the physician continues to assume the more powerful position in the relationship. This power imbalance is not merely

implied however, because ultimately the physician has the authority to refuse to carry out the patient's preferred treatment option (Charles et al., 1999).

Yet the requirement to provide appropriate information to patients, which is an essential component of the process of informed consent and helps to facilitate the participation of patients, has not always been central to treatment decision-making, only having become accepted as standard practice in the UK and the US as the nature of decision-making has evolved over the latter half of the 20th century (Entwistle and Watt, 2006). Furthermore, patient participation is an ambiguous and vague concept, one that is over-simplified in policy, and yet argued to be a realistic goal that is not only morally and ethically sound, but one that both patients and the medical profession desire (Entwistle and Watt, 2006).

The process of informed consent underpins the model of informed choice, which for Jonsen et al. (2006), "is the practical application of respect for the patient's autonomy" (p.54), and has become the crux of treatment decision-making, enshrined in both case law, and in the codes of conduct and guidance registered medical professionals must adhere to (Royal College of Paediatrics and Child Health, 2002; General Medical Council, 2008; Nursing and Midwifery Council, 2008).

Although "there is no English statute setting out the general principles of consent" (DH, 2001a, p.29), any failure by a professional to obtain informed consent before proceeding with a proposed treatment or intervention is both illegal and unethical, and can result in serious penalties for the professional involved (DH, 2001a; Jonsen et al., 2006).

In order for the medical professional to obtain the informed consent of a patient, at the very least a one-way exchange of information from physician to patient about the benefits and risks of a proposed treatment, intervention or test, is essential. However, Jonsen et al. (2006) argue that professionals must avoid viewing consent "as a mechanical recitation of facts or a pro forma signature on a piece of paper" (p.55), which they argue "reveals a fundamental misunderstanding of informed

consent” (p.55). They stress, that for consent to be informed, three key principles underpinning the process must be achieved:

- consent must be made by a *competent* patient
- consent must be *informed*
- consent must be *voluntary*.

These essential features of informed consent are emphasised by Buchanan and Brock (1990), who argue that “the informed consent doctrine requires the *free* and *informed* consent of a *competent* patient to medical procedures that are to be performed” (p.26). More recently, the Department of Health, recognising the legal and ethical status of informed consent in modern medical practice, produced a reference guide to consent for all health care professionals, in which they argue that for consent to be valid, “it must be given *voluntarily* by an appropriately *informed* person (the patient or where relevant someone with parental responsibility for a patient under the age of 18) who has the *capacity* to consent to the intervention in question” (DH, 2001a, p.4).

To have decision-making capacity, a patient “must be able to comprehend and retain information material to the decision, especially as to the consequences of having or not having the intervention in question, and must be able to use and weigh this information in the decision-making process” (DH, 2001a, p.4). The Mental Capacity Act 2005 breaks down this process into four key elements, the final one, to be able to communicate the decision (verbal or otherwise), dependent on the patient being able to meet the first three:

- to be able to understand relevant information about the nature, reason, and likely effects of choosing between the options available, including the decision to take none of the available options
- to be able to retain the information long enough to use it for making a decision
- to be able to use and weigh the information provided in the decision-making process (referred to as ‘logic’ in decision theory, and an ability beginning to

develop in children from the age of about seven (Friedman Ross, 1998))
(Department of Constitutional Affairs (DCA), 2007).

Where patients have some capacity to participate, and this would be the case for children and young people who have developing capacity for decision-making, it is possible that the patient's ability to process information, in other words to become informed, is impaired or under-developed, or that their vulnerability and limited capacity makes them at risk of coercion or manipulation, therefore posing a challenge to the principle of informed voluntary consent. As Buchanan and Brock (1990) argue, "if the decision is not voluntary, but instead coerced or manipulated, it will likely serve another's ends or another's views" (p.26). They also point out that "if the appropriate information is not provided in a form the patient can understand, the patient will not be able to ascertain how available alternatives might serve his or her aims" (p.26).

What is and is not appropriate information is a controversial and potentially litigious element of informed consent, and Jonsen et al. (2006) point out that in the past, "what a reasonable and prudent physician would tell a patient" (p.56) was perceived as a good enough standard. However, as the principles of self-determination and patient autonomy have gained in prominence, a more patient centred standard, referred to as the 'reasonable-patient' standard, and defined as the "information reasonable patients need to know to make rational decisions" (p.56), has grown to replace it.

Although the models of informed choice and shared decision-making have at their core the process of informed consent, they are each underpinned by a distinct rationale, offer a different vision of the essential characteristics of treatment decision-making, and envisage different roles for both the physician and patient in the decision-making process. The models also differ in how they view the process of information exchange, and the assumptions regarding the patient's ability to understand and use information for decision-making are quite different.

While the legal and ethical requirements of the physician to obtain informed consent from the patient goes some way to shifting control to the patient, the patient is nevertheless dependent on the physician to “provide truthful information, to maintain competence in their area of expertise, and to consult others when their knowledge or skills are lacking” (Emanuel and Emanuel, 1992, p.2221). In recognition of this challenge for the models of informed choice and shared decision-making, and the implications for professionals who fail to obtain informed consent from the patient, there has been a growth in the development of treatment decision aids, designed to “inform patients about available treatment options and their benefits and risks, and to structure the decision-making process in order to encourage patients to express their treatment preferences” (Charles et al., 2005, p.114).

3.3 The Concept of Shared Decision-Making

Acknowledging the unhelpful dichotomy between paternalism and informed decision-making, and in response to growing concerns about the model of informed choice, which reduces the role of clinician to one of information provider and places huge expectations on the patient to become informed and take ultimate responsibility for treatment decisions, the model of shared decision-making “merges these models into one where patients and physicians participate equally in all stages of decision-making” (Flynn et al., 2006, p.1159-60). For Edwards and Elwyn (2006), “as neither party is fully able to derive and use the knowledge and experience or personal values and goals of the other, shared decision-making offers a possible middle-ground with each party contributing to a decision” (p.308).

While several authors offer a clearly defined conceptualisation of shared decision-making (Charles et al., 1999; Coulter, 1999; Towle and Godolphin, 1999; Elwyn et al., 2000), Moumjid et al. (2007), following a review of the literature in this area, revealed that a third of the papers referring to shared decision-making fail to provide a definition or to reference a source for the model. They argue, therefore, that there remains much ambiguity and confusion surrounding the concept of

shared decision-making, particularly when some authors employ the terms of informed and shared decision-making interchangeably, failing to identify them as distinct models with different characteristics and underpinning assumptions (Moumjid et al., 2007). To complicate matters further, Towle and Godolphin's (1999) conceptualisation of this model of treatment decision-making, while useful because of its consideration of the physician and patient competencies required for effective decision-making, is referred to as 'informed shared decision-making'.

For Moumjid et al. (2007), Towle and Godolphin's (1999) model, along with the models offered by Charles et al. (1999) and Coulter (1999), not only provide a clear definition of the meaning of shared decision-making, but distinguish it from the process of informed choice by emphasising the requirement that "patients and health professionals share both the process of decision-making and ownership of the decision made" (Coulter (1999) referenced in Moumjid et al., 2007). They are also referenced by a third of the authors referring to shared decision-making in Moumjid et al.'s review, and share the same overall view of shared decision-making as a process in which the physician and patient work together, as partners, using and discussing the information and preferences held by both parties to make an informed decision.

It is argued here that the model of Charles et al. (1999) offers the most useful framework of the process of shared decision-making in view of the fact that it incorporates the paternalistic and informed perspectives within the overall framework, and establishes different components, or analytic stages, in the decision-making process, therefore providing an insight into what Edwards and Elwyn (2006) refer to as the black box of treatment decision-making. For Flynn et al. (2006), these elements of the model help to "accommodate situations where patients want to participate to different degrees in different stages" (p.1160). Charles et al. (1999) in fact argue, that when compared to their original conception of shared decision-making (Charles et al., 1997), the "revised and updated framework ... is more flexible and incorporates a more dynamic perspective on treatment decision-making" (p.658).

While the key features of shared decision-making identified in their original conception are still applicable to the revised model, they are now offered as guiding principles rather than “minimum or necessary criteria for classifying a physician-patient decision-making interaction as shared” (Charles et al., 1997, p.685). These principles state that shared decision-making involves the *participation* of at least two participants (the physician and patient), that both parties take steps to participate in the process of decision-making by *sharing information* and *expressing treatment preferences*, and that a *treatment decision is made and agreed upon* by both parties.

From reviewing the conceptual literature concerned with treatment decision-making, the revised model developed by Charles et al. (1999) was selected to inform the current research, and is presented on the following page (p.69). Acknowledging concerns about the use of typologies and models through which to understand how decisions are made in practice, and taking into account the lack of empirical evidence to support the models presented in the literature, the continuum of patient control discussed by Thompson (2007) has been incorporated within the framework. This serves to illustrate how the framework integrates the diversity in decision-making and the varying degrees of patient control across the models included. As Thompson (2007) points out, patients “may wish to be involved at different levels in relation to different circumstances and it may change over time for the same person in the same context” (p.1308). It is therefore essential that the framework underpinning the current work reflects this complexity and fluidity.

The framework identifies the analytic stages in the process as information exchange, deliberation, and decisional control (the term they employ is ‘deciding on treatment to implement’), further categorising the exchange of information by flow, direction, type and amount. Although the phases are represented as separate components in the model, Charles et al. (1999) rightly acknowledge that it may well resemble a more iterative process. Before discussing the analytic stages, it is worth noting that this is not an exhaustive review of decision-making models, and other models can be placed on the continuum. For example, Charles et al. (1999) refer to

the physician-as-agent model in which the patient’s values are communicated during the decision-making process but decisional authority remains with the physician, there positioned between that of shared decision-making and paternalism.

Table 1 – Framework of Treatment Decision-Making Models

Adapted from Charles et al. (1999) and Thompson (2007)

ANALYTICAL STAGES	Decisional Control Continuum		
	Low level of patient power		High level of patient power
	←	→	
	Paternalistic	Shared	Informed
Information exchange			
<i>Flow</i>	<i>One-way</i>	<i>Two-way</i>	<i>One-way</i>
<i>Direction</i>	<i>Physician to patient</i>	<i>Physician to patient</i>	<i>Physician to patient</i>
<i>Type</i>	<i>Medical</i>	<i>Medical and personal</i>	<i>Medical</i>
<i>Amount</i>	<i>Legally required</i>	<i>All relevant for decision</i>	<i>All relevant for decision</i>
Deliberation	<i>Physician alone or with other physicians</i>	<i>Physician and patient (plus potential others)</i>	<i>Patient (plus potential others)</i>
Decisional control	<i>Physician</i>	<i>Physician and patient</i>	<i>Patient</i>

Emanuel and Emanuel (1992) include in their review the deliberative and interpretive models, which resemble the model of informed choice in that the physician provides information to the patient regarding the risks and benefits of the proposed intervention, and the patient retains decisional control. The interpretive model, however, recognises that the patient’s values “are not necessarily fixed and known to the patient” (p.2222), and the physician has an additional role to work with the patient to help them clarify and understand their values within the context of the medical situation, therefore positioned on the continuum between that of shared decision-making and informed choice. The deliberative model extends the physician’s role further, incorporating their professional expertise and preferences

regarding treatment in order to help the patient “judge the worthiness and importance of health-related values” (p.2222), thus gaining itself a similar position but one closer to that of shared decision-making than the interpretive model.

3.3.1 Information Exchange

Charles et al. (1999) state that “information exchange refers to the type and amount of information exchanged between physician and patient and whether information flow is one or two way” (p.654). For shared decision-making, Charles et al. (1997) argue that “there needs to be a two-way exchange not only of information but also of treatment preferences” (Charles et al., 1997, p.685), which contrasts to the models of paternalism and informed choice in which the patient’s knowledge and preferences are not made explicit. In fact, a key feature of information exchange in the model of shared decision-making is that the narrow focus in the models of informed choice and medical paternalism, on the risks and benefits of the proposed treatments, is broadened to include other types of information patients may use in the decision-making process.

In fact, because the process of information exchange is a central component of the shared decision-making, rather than a requirement of the physician to provide relevant information in order to obtain informed consent, greater attention is paid to the information needs of the patient. As all information relevant to the decision is a feature in this model, including that which is relevant for the patient as well as the physician, it is likely to include a range of different types and forms of information, from different sources, and the amount involved will vary considerably by patient, and by decision context.

As Charles et al. (1999) point out, there are no outer boundaries within this model, although they note that “the amount of information exchanged will be influenced by time and money constraints” (p.655). Charles et al. (1999) categorise the information as follows:

- Physician to patient – natural history of illness, benefits and risks (side effects) of treatment options, potential effects on psycho-social wellbeing as well as

physical health, recommended option, community resources and support, sources of other information the patient could access

- Patient to physician – experience of illness, lifestyle (values, quality of life) and social context (family and work life), beliefs and fears about illness, knowledge of illness and treatment options (obtained from other sources such as patient support groups and the internet)

In addition, Charles et al. (1999) recommend that as part of the process of exchange, “either the physician, the patient, or both may exchange preferences regarding their own and each other’s role in the decision-making process” (p.654), although Elwyn et al. (2000) argue that this should take place following the exchange of information in order for the patient to have a better idea, based on the complexity and seriousness of the decision, of their preferences for participation and decisional control. While recognising the importance of this aspect of decision-making, Charles et al. (1999) fail to address whether a decision in which the roles for participation are negotiated constitutes a shared approach when those preferences result in a scenario that resembles paternalism or informed decision-making. This failure to incorporate the negotiation of roles within the broader framework is a weakness, especially as for Degner (1998) it is this aspect of decision-making, as opposed to participation itself, which gives the patient control over the process.

3.3.2 Deliberation

Deliberation “refers to the process of expressing and discussing treatment preferences” (Charles et al., 1999, p.656). The exchange of information alone, Charles et al. (1999) argue, does not constitute a shared approach. Although in the paternalistic and informed choice models a one-sided deliberation may take place, the physician weighing up information on behalf of the patient in the former, and the patient weighing up information, perhaps through discussion with significant others in the latter, the key feature of deliberation in shared decision-making is its ‘interactional nature’. In fact, Ruland (2005) argues that the deliberative feature of

shared decision-making is particularly useful when “outcomes are uncertain, when clinicians need to pay attention to individual circumstances, when patients’ values are variable and unknown, and when a decision involves making value judgments” (p.70).

Charles et al. (1999) argue that for deliberation to have meaning, “both physicians and patients have to perceive that there are treatment options” (p.656). This is known as ‘clinical equipoise’, re-conceptualised as ‘professional equipoise’ by Elwyn et al. (2000), and referring to those decision-making situations in which the physician has “no clear preference about the treatment choice to make” (p.895). They agree with Charles et al. (1999), that “this is where shared decision-making is most feasible” (p.895), and while it usually means there is more than one viable treatment, there are cases in which both agreeing, and not agreeing, to a single proposed intervention constitute two reasonable choices, whether due to trade-offs between options, prognostic uncertainty, or the subjective and quality of life factors involved.

Where exchanged preferences for treatment diverge, the stage of deliberation may well include a process of negotiation in order to reach an agreement, and ultimately share the decision. However, while Charles et al. (1999) recommend that negotiation takes place between ‘equal partners’, this process is hindered by the “power, status and informational asymmetry between physician and patient” (p.659), as well as other well-known divisions of inequality such as gender, ethnicity, education, income and culture. What’s more, in cases where deliberation and negotiation is extended due to differing views and preferences, Charles et al. (1999) admit that it can “make the process more cumbersome and time consuming” (p.656). There is also a danger within this approach that an agreement cannot be made, which in the extreme scenario can result in the physician refusing to implement the patient’s preferred treatment option.

3.3.3 Decisional Control

Choosing a treatment to implement is the final part in the decision-making process, and must follow that of information exchange and deliberation if the process of informed consent is to be achieved. In shared decision-making, both the patient and the physician, “through the deliberation process, work towards reaching an agreement and both parties have an investment in the ultimate decision made” (p.658). This contrasts with the model of paternalism in which the physician assumes decisional control, and the model of informed choice in which the patient has control over the final decision. The advantage of such an approach is that the overall responsibility for the treatment decision is shared between the patient and physician, which Elwyn et al. (2000) argue has the potential to facilitate participation when patients are reluctant to assume decisional control, particularly when “choices are difficult and the issues painful” (p.896).

3.3.4 Guidelines for shared decision-making

A number of authors, recognising the challenge physicians face in facilitating a more collaborative approach to treatment decision-making, have incorporated within the writings on shared decision-making, guidelines and competency frameworks for health care professionals. These range from competency frameworks for patients and physicians (Towle and Godolphin, 1999; Towle et al., 2006), checklists for the process of information exchange (Coulter et al., 1999), and guidelines for the process of shared decision-making (Elwyn et al., 2000). Many of these are now grounded in empirical work looking at the process of shared decision-making in practice, including the suggested skills and steps proposed by Elwyn et al. (2000):

- Develop a partnership with the patient
- Establish or review the patient’s preference for information, e.g. amount and format
- Establish or review the patient’s preferences for role in decision-making
- Ascertain and respond to patients’ ideas, concerns, and expectations

- Identify choices and evaluate the research evidence in relation to the individual patient
- Present (or direct to) evidence, taking into account the above steps, and help the patient reflect upon and assess the impact of alternative decisions with regard to their values and lifestyles
- Make or negotiate a decision in partnership, manage conflict
- Agree upon an action plan and complete arrangements for follow-up
(adapted from p.893)

This step-by-step process is useful because it demonstrates some of the key elements that in the model developed by Charles et al. (1999) occur at the periphery, including the process of identifying treatment options in relation to the individual patient, which although having a bearing on the options ultimately offered to the patients, is excluded from the physician-patient encounter. The importance of follow-up is also identified as a crucial aspect of decision-making, and provides the patient with an opportunity to clarify any misunderstandings and seek additional information they need after having time to reflect on the decision.

3.3.5 Preferences for shared decision-making

As the research around participation in decision-making grows, it has become apparent that while in most cases patients wish to receive information about a given treatment, intervention or test, they do not necessarily wish to be involved in the decision-making process, or to assume decisional control (Flynn et al., 2006). Furthermore, while there is a growing consensus that patients should be encouraged to participate in decision-making (Thompson, 2007), it continues to be argued by some authors that medical paternalism is still preferred for decisions in which there is only one clear treatment option, and essential “during emergencies when the time taken to obtain informed consent might irreversibly harm the patient” (Emanuel and Emanuel, 1992, p.2224), drawing attention to the conflict between patient autonomy and the physician’s obligation to prioritise the patient’s health over the patient’s values and preferences.

There is a pervading theme across the literature concerned with health care decision-making that patients should be actively encouraged to participate in and assume control of decisions regarding their care and treatment, culminating in the ideal models of informed and shared decision-making. But is this the reality of decision-making in practice? And should it be the case that patients who prefer their physicians to take responsibility for their treatment decisions are persuaded to do otherwise? These are some of the questions now being addressed in the research, which, in acknowledging the assumptions regarding the supposed benefits of participation for patients, have begun to explore patient preferences for decision-making, and the congruence between these and the roles patients actually assume in decision-making situations.

The findings of these studies are illuminating because while some studies have found social divisions of gender, education, age, and income to have an effect on participation, other studies contradict these findings, revealing the importance of the decision context with regards to preferences for, and participation in decision-making, also highlighted by Elwyn et al. (2000) and Bekker et al. (1999). So while it is true that patients want to be 'informed', and there is a strong desire for information, they do not necessarily want to assume decisional control. Furthermore, while some patients wish to discuss their treatment options with their physician, others do not. Viewing preferences for the process of decision-making overall is therefore misleading, and reveals the utility of Charles et al.'s (1999) revised model of shared decision-making, which breaks down the components of the decision-making process into three distinct phases.

Regardless of patients' preferences for participation in decision-making, Charles et al. (1997) remind us that it 'takes two to tango', in other words the clinician must also have a preference for a more shared decision-making approach. However, Elwyn et al. (1999), who conducted focus groups with general practice registrars, in which their views about shared decision-making were elicited, found diversity in attitudes towards what they perceived to be a novel approach to clinical decision-making with some practitioners still fiercely protective of the paternalistic model.

Among the registrars who could see the benefits of such an approach, which they argued was more suited to situations involving professional 'equipoise', there were concerns about the ability of patients to understand medical data involving risks and probabilities, and their own ability, as generalists rather than specialists, to provide information of the depth and accuracy required. In addition, time constraints, and the lack of communication skills required to facilitate shared decision-making, were highlighted as a potential barrier.

When asked about their current practice, many of the registrars spoke about presenting data in a way that 'steered' patients towards the recommended treatment option, and very few admitted to seeking patient preferences for participation, although there was "an underlying assumption that most patients do want to be involved" (Elwyn et al., 1999, p.755). A more recent study by Burges Watson et al. (2008), also consulting general practitioners on the topic of shared decision-making, found that while practitioners acknowledged the growing role of patients in decision-making, there were two quite distinct approaches to the practice of shared decision-making. The first, patient-centred, was promoted by practitioners who value the principles of equality and patient autonomy, consistent with the models proposed in the literature. However, the second, practitioner-centred, remained grounded in the expertise of the medical professional, and for some practitioners, veered towards paternalism.

The research regarding physicians' preferences is beginning to reveal a general consensus that involving patients in decision-making is a desirable goal (Gravel et al., 2006), and a recent survey by Murray et al. (2007) suggests that around three quarters of physicians prefer a shared decision-making approach. However, the studies reveal a number of barriers to implementing it in practice, including time constraints, communication barriers, and the difficulty of eliciting patient preferences (Edwards and Elwyn, 2006; Gravel et al., 2006; Towle et al., 2006; Murray et al., 2007). Moreover, the type of physician-patient relationship required for shared decision-making, one of partnership, is fundamentally different to the way in which patients, and physicians, expect it to be, given the predominance of

paternalism throughout many generations. Towle et al. (2006) argue, therefore, that is a “need to change well-established patterns of communication with patients” (p.331).

There is very little research about the preferences of other health care professional groups for sharing decision-making, and this is perhaps because the conceptual literature concerned with making treatment decisions has developed around the doctor patient relationship, and the shift from medical paternalism towards shared and informed decision-making is also based on this decision dyad (Gravel et al., 2006). However, within the literature reviewed in Chapter Two there is a general agreement that a multi-disciplinary and shared approach to supporting families has the potential to improve decision-making for children and young people with life-limiting conditions (Todd et al., 2005; Wright et al., 2009), and this will be discussed in more detail in Section 3.4.2 which addresses the relevance of shared decision-making in the paediatric setting.

3.4 Treatment Decision-Making for Children and Young People

One of the major limitations in the models of medical decision-making – including the preferred model of shared decision-making now advocated in policy and practice is the focus on the patient-physician dyad. This bias is also evident in the research concerned with preferences for and participation in decision-making, whether or not underpinned by a conceptual model. However, this is not only unrealistic for complex decisions involving an adult patient as it is likely that a family member or other key professional may be involved in the decision process, but it does not reflect in any way the triadic nature of paediatric decision-making, which is complicated further due to the unique legal status of parents, children and medical professionals in the decision-making process.

In addition, for children with complex health care needs and those requiring palliative care, many decisions will occur over a period of time, and extend beyond the triad of patient child or adolescent, parents or legal guardians, and the referring

health care professional. For example, it is increasingly the case that a multi-disciplinary health care team consisting of a range of health care and other professionals supporting the family is involved in the on-going and complex decisions regarding their condition (Sourkes et al., 2005). It is also quite possible that other family members, carers, and family friends may be involved in discussions about planned interventions, especially those involving a fair degree of uncertainty with regards to a child's quality of life. Although the concept of family-centred care provides a broader framework in recognition of this complexity, within which shared decision-making is advocated (Hutchfield, 1999; Shields et al., 2006), there is, as yet, no model of decision-making that adequately represents this process.

In fact, very few authors concerned with treatment decision-making for children refer to the model of shared decision-making although Bauchner (2001) does argue that "shared decision-making is an important aspect of paediatric care" (p.246), admitting that it is still quite a new concept and suggesting that shared decision-making may not be that different from family-centred care. While the nature of decision-making for children has in the past reflected the paternalistic model, with parents assuming the passive position in the relationship and accepting the decisions made by their child's physician, they too, like adult patients, have gained power in the medical situation, partly because of the requirement upon health professionals to obtain informed consent, which for the child patient needs to come from a parent or legal guardian, the person now recognised in law as being responsible for the child's upbringing (Children Act 1989).

Similarly, children, who were once invisible in the process of decision-making, are now increasingly being encouraged to participate in decisions regarding their treatment and care, reflecting a broader shift towards acknowledging the rights of children, including the right to self-determination and autonomy associated with the process of informed consent (Hallström and Elander, 2005; Moore and Kirk, 2010). This follows a long history in the UK of viewing children as an inseparable, and silent part of the family unit, and childhood as a period of 'becoming', requiring

protection, socialisation and education as the child develops into an adult and future citizen (Hill and Tisdall, 1997). As a result, and influenced by the view that family life is “an area in which government should not intrude (Hill, 2003, p.289), children as individuals have had virtually “no social policy identity” (Daniel and Ivatts, 1998, p.7), the government tending instead to adopt a limited role of protecting the welfare of vulnerable children.

However, the 1990s were “a time for adults to re-examine their perceptions of children and attitudes towards them” (Hill and Tisdall, 1997, p.2), resulting in a cultural and political shift towards viewing childhood, a term now generally acknowledged as “a socially constructed classification” (Daniel and Ivatts, 1998, p.2), as a process of ‘being’, as well as ‘becoming’. In other words, children were beginning to be seen, for the first time, as “individuals in their own right and not merely, or even mainly, as future adults” (p.2). This was accompanied by the growing recognition that from very early on children “contribute actively to the creation of the social worlds in which they live, both individually and collectively” (Daniel and Ivatts, 1998, p.1).

3.4.1 The legal and ethical context

This idea that children are ‘social actors’ with the capacity to influence as well as to be influenced by their social environment (Prout, 2002, p.67) has given rise to increasing pressure for the government to give children a public voice. As Hill and Tisdall (1997) point out, children’s lives are shaped by social and economic policy, and therefore children should have the opportunity to speak up and be heard in the public arena. In December 1991, the foundations for this goal were laid as the United Nations Convention for the Rights of the Child, an international agreement acknowledging children as active citizens with distinct needs and rights, was ratified in the UK. With regards to decision-making, Articles 12 and 13 of the Convention emphasise children’s rights to both express their views concerning decisions affecting them, and have them weighted according to age and maturity; and to

receive information in an appropriate form in recognition of their right to freedom of expression (United Nations, 1989).

These rights, acknowledged in the Children Act 1989 with regards to the welfare of vulnerable children, have also been made explicit in the area of treatment decision-making. In 2004, the National Service Framework (NSF) for Children, Young People and Maternity Services (DH, 2004a), setting out the government's ten year programme "to stimulate long-term and sustained improvement in children's health" (p.8), argues that:

"Parents and their children should be central to the process of decision-making about their care, and receive appropriate information to exercise choice. They need to be fully informed and provided with information about the nature of the illness, different interventions and treatment options available to them and relevant support groups and voluntary organisations that might help them" (Standard 6 of the National Service Framework; DH, 2004b, p.10).

This transformation in how the role of the child and adolescent patient is viewed for decision-making is also evident in the changing conceptualisation within medical ethics, and in law, to the process of informed consent for children and young people which, as a pre-requisite to both the models of shared and informed decision-making, needs elaboration and expansion if it is to become meaningful in the paediatric setting.

It has already been summarised that for the child patient, a parent or legal guardian acts as the decision-maker. It is also the case that the standard principles of informed consent – that a decision must be informed, that a decision must be voluntary, and that a decision must be made by a competent patient – still apply for this situation although it is the parent rather than incompetent child who must have decision-making capacity (DH, 2001b). In addition, the person with 'parental responsibility', generally a parent or legal guardian, although in some instances a Local Authority or the courts (according to the definition in The Children Act 1989),

must base their decision “according to the “welfare principle”, in other words “that the child’s “welfare” or “best interests” must be paramount” (DH, 2001b, p.18). For children where more than one person has ‘parental responsibility’, consent given by one person is valid “even if another person with parental responsibility withholds consent (p.18).

Although the age at which a young person can legally consent or refuse treatment, assuming they have decision-making capacity, is sixteen, there is some ambiguity surrounding young people age sixteen and seventeen, partly because previous legislation puts the age of majority (Family Law Reform Act 1969), and of full consent (Children Act 1989), at 18 (Wright et al., 2009). For young people age sixteen and seventeen who are viewed in general terms as having capacity for decision-making, and who can give informed consent in their own right, there are certain cases in which their consent, and in particular their refusal for treatment can be overridden by a person with parental responsibility or by the court, under the ‘best interests’ standard, although the wishes of the young person will be taken into consideration (DH, 2001b, p.16). What’s more, advance decisions (known as advance directives) to refuse treatment, can only be made by patients aged 18 and over (DCA, 2007).

The issue of competency in fact, poses a major challenge for the paediatric setting because of the growing recognition that decision-making capacity depends on a unique combination of cognitive ability, personal experience, and maturity. In the past, capacity has been measured solely by a child’s age, which although in general can predict cognitive development and maturity, is now agreed to be oversimplified, and a rather crude measure of competence (Friedman Ross, 1998). Therefore, although medical decisions for children under the age of 16 continue to be “authorized by the usual surrogates, namely, the parents of the child or, in unusual circumstances, by other parties authorized by the law” (Jonsen et al., 2006, p.92), the given age at which children are deemed to become competent decision makers is no longer used in cases assessing a young person’s capacity, which instead employ the concept of *Gillick* competence (DH, 2001b).

For children under the age of 16, and for young people age 16 and 17 whose capacity is questionable, *Gillick* competence argues that “children who have sufficient understanding and intelligence to enable them to understand fully what is involved in a proposed intervention will also have the capacity to consent to that intervention” (DH, 2001a, p.16). As for adults, capacity is viewed as decision-specific, and competence is therefore assessed on the child’s developing maturity and capacity as well as the seriousness and complexity of the decision (DH, 2001b). Wright et al. (2009) argue that to meet the criteria for *Gillick* competence, a young person must “understand and appraise the nature, purpose and implications of treatment; any risks there might be and any alternative courses of action and their consequences” (p.239). They add that in practice, “it is necessary to consider emotional maturity, intellectual capacity and psychological state” (p.239).

Competency alone, however, is not enough to make an informed decision, and special attention must be paid to both the process of information exchange for children and young people, and to the dynamics of the doctor-child, parent-child relationship in order for consent to be *voluntary* and *informed* (DCA, 2007).

Professionals therefore have a duty to ensure suitable information, employing appropriate language and terminology, and available in a child-friendly format, is provided in order for competent children to weigh the benefits and risks of the proposed treatment (DH, 2001b). Professionals need to be aware not only of the influence parents, carers, or significant others, can have on children and young people in decision-making situations, but also of the inherent power imbalance between themselves – both as an adult and as a professional – and the child patient (DH, 2001a).

A final issue in the process of informed consent for children relates to the requirement upon parents to make decisions for their child using the ‘best interests’ standard, a standard increasingly upheld as children have come to be viewed as individuals in their own right, and not simply as the property of their parents. McCabe (1996) points out, that while parents continue to be viewed as “the most motivated and capable people to act in their children’s best interests” (p.507), there

are situations in which there is disagreement about the treatment or intervention that meets this standard (Whitney et al., 2006). McCabe (1996) suggests that “it is in highly ambiguous decision-making situations, which are emotionally burdened, that parents’ capacities may be compromised” (p.507). Furthermore, in situations involving subjective values including issues around quality life, disagreement can be found within the same family or professional team (Wright et al., 2009).

For Friedman Ross (1998), conflict can also arise in situations where the parent’s dual obligation to act in the best interests of their child *and* the wider family unit, results in what the professional, whose primary obligation is to act in the best interests of the patient, views as the wrong choice. However, from the parent’s perspective, if the family unit in which the child lives is adversely affected by a decision made in the best interests of the child, then the child’s life may also be adversely affected. Although the premise of family-centred care acknowledges this problem, particularly for the family of a child with complex health care needs whose existence and functioning may well be impeded due to the child’s complex care regime, the physician, underpinned by the primary obligation to act in the patient’s best interests, may not consider this factor to be of primary importance in the decision-making process.

Whitney et al. (2006) argue that in circumstances where there are two ‘clinically reasonable choices’ which involve the weighing of subjective and value-based information, “the family, with its deeper understanding of the child’s nature and preferences, is better positioned to take the lead” (p.160). McCabe (1996) adds that in these situations, “children should be afforded the opportunity for greater involvement” (p.507), although she notes that the weight accorded to the child’s views should be made with their developing competence in mind. In fact, there is a growing consensus, in recognition of the rights afforded to children, including the rights to self-determination, and autonomy, that the child’s wishes and views should be incorporated in the decision-making process regardless of their competence. For Hallström and Elander (2005), doing so not only “demonstrates respect for children and may provide opportunities for further decision-making

capabilities” (p.225), but it also has the potential to improve communication between those involved. McCabe (1996) adds that enabling children to participate may enhance treatment adherence and cooperation by incorporating the child’s wishes within the decision process, and promote a sense of control for the child or young person.

With this in mind, the American Academy of Pediatrics (AAP) (1995) argues that rather than informed consent, which should be applied only to “patients who have appropriate decisional capacity and legal empowerment” (p.314), the concepts of informed (parental) permission and (child) assent/dissent should be applied in decisions involving the incompetent child. Hallström and Elander (2005) define assent as “an interactive process between the child and adults, and requires that the child has been informed about what is going to happen and has agreed to the procedure” (p.227). Dissent, on the other hand, “involves a difference of opinion and a lack of agreement between the child and the other parties” (p.227). Hallström and Elander (2005) argue that, while dissent can be over-ruled by the adult with ‘parental responsibility’, a child’s dissent, given they understand the information provided, should be taken seriously by those involved.

By incorporating the concept of assent within the process of decision-making, attention is paid to both the child’s developing capacities for decision-making, and their right to autonomy. According to the AAP (1995), assent involves more than a child simply agreeing to a procedure without understanding what is involved, arguing that children are empowered “to the extent of their capacity” (p.314). However, the Canadian Paediatric Society (2004) point out that, while assent offers the incompetent child “some authority over their own health care” (p.101), there is a danger that children are “given the impression that they have more control than they do” (p.101), a concern also discussed by Hallström and Elander (2005) who view assent as the lowest level of participation because ultimately others will make the decision, even though the child may have some choices.

3.4.2 Shared decision-making in paediatrics

Despite the changing nature of informed consent for children and young people, Whitstone (2004) reminds us that for treatment decisions, which continue to be made on the basis of 'informed consent', "the authority still lies in the hands of parents or legal guardians who give their permission for treatment to take place". Although shared decision-making is promoted as an ideal type and actively encouraged in the National Service Framework for Children (DH, 2004), very little is known about how children and young people are involved in treatment decision-making and there are currently no conceptual models that address this particular process (Stewart et al., 2005).

There are some existing broader models of child participation that explore the different ways in which children can participate in the development of services and policy, and aim to establish the status of children as 'citizens' (Hart, 1996; Shier, 2001). Drawing on the relevance of this work for treatment decision-making it is easy to see why children might be offered a tokenistic level of participation, which on the surface meets the requirements set out in the National Service Framework and may even reflect the nature of child assent, but at the same time avoids the potential for conflict between the parties involved because the level of participation is not meaningful (Shier, 2001).

Franklin and Sloper (2009) suggest that in fact children are becoming more involved in decision-making than has been possible in the past, and there are some recent studies to suggest this is the case (Knopf et al., 2008; Bluebond-Langner et al., 2010; Coyne and Gallagher, 2011; Moore and Kirk, 2010). However, this new field of research provides conflicting evidence about the preferences young people have for treatment decision-making, and Knopf et al. (2008) found that among the 82 adolescents taking part in their research, 44% preferred passive decision-making (medical paternalism), 37% preferred a shared approach, and 17% preferred active (informed) decision-making. In addition, whilst there is a general assumption that parents promote the idea of child participation, there is a lack of evidence to

support this and Knopf et al. (2008) found that some parents prefer to assume decisional control in the guise of protecting their child from a potentially stressful situation.

Given the complexity of decisions about technological support and the difficult role parents assume as decision-makers for their child, this is quite possible, and in supporting families through decisions about technological support Gauderer (2002) emphasises the “importance of an inclusive multidisciplinary teamwork in the indication and the management process” (p.109). Ali et al. (2005) and McHattie (2005) also recommend a multidisciplinary approach, and Rouse et al. (2002), who interviewed both parents and professionals as part of a small qualitative study, revealed an overall consensus “that a multidisciplinary approach was vital for parents’ understanding of the issues involved” (p.126). However, like participation, multi-disciplinary working is fraught with conceptual differences and practical challenges, and again there is little empirical research to support the argument that decision-making is better when it is conducted in this context.

As well as a multi-disciplinary approach, Samuel and Boit (2007) stress the importance of ‘planning’ which, while not always possible for children who present as life-threatening at the time of a decision, can otherwise provide the family time to think about what the intervention means for them and for their child, and the long-term consequences involved. For Ali et al. (2005), clinicians involved in complex treatment decisions must be able to adopt a flexible approach and continually assess how parents and young people wish to be involved in the decision-making process, and how objectively they are able to weigh up the risks and benefits of interventions that involve clinical uncertainty. They put forward the following questions which they argue to be crucial for assessing the information and support required to make informed decisions:

- Are patients and surrogates well informed enough to make a reasonable decision?
- How much advice should clinicians offer without being paternalistic?

This issue about how much information to provide patients and their families is also raised in the literature regarding shared decision-making, with physicians identifying concerns about the ability of patients to process information about the benefits and risks of interventions. Ali et al. (2005) add that clinicians are also faced with the dilemma that too much information may overwhelm families yet it is unethical to withhold any relevant information. This presents the clinician with an ethical dilemma about how much information to provide each family, and whether it is ethical at all to vary the information based on an assessment of their desire and capacity for information, and their ability to weigh up the benefits and drawbacks for their child.

A fundamental feature of the decision-making process in any clinical encounter is 'communication' between clinician and patient, and in the case of paediatrics the triad of clinician, child / adolescent patient and parents. To discuss preferences for participation in decision-making and the needs families might have for information and support, open and honest communication must take place, an approach recommended by the Royal College of Paediatrics and Child Health (2004) who argue that "open and timely communication between the young patient, family and members of the Health Care Team is central to informed and ethical decision-making" (p.23).

Although the referring clinician is likely to take the lead in decisions about technological support, Todd et al. (2005) advocate a role for nurses in decisions about technological support, and found them to have a great deal of knowledge and experience that was often under-utilized in the decision-making process. Furthermore, they observed that decision-makers were often confused and dissatisfied with the information they had received, and spoke openly about this with the nursing staff. Todd et al. (2005) argue that "the close and trusting relationships which nurses often establish while caring for patients with extended illnesses places them in an effective position to help these patients and their families in making such decisions" (p.188). Rollins (2006) too notes that "a well-informed nurse could play a central role in dispelling myths surrounding tube

feeding” (p.22), reflecting Kirk’s (1999) argument, in her discussion about the changing role of community-based nurses, that “being an information resource for parents will become increasingly relevant and important” (p.393).

3.5 Using Shared Decision-Making as a Framework to Explore the Decision Process

The research concerned with treatment decision-making in the paediatric setting has a tendency to focus on the participation of young people, and very few studies concern themselves with the actual process of decision-making (Stewart et al., 2005). Although there is general support for a shared approach within the literature and growing recognition of the multi-disciplinary nature of paediatric palliative care (Craft and Killen, 2007), there continues to be an evidence gap into how treatment decisions are made for children and young people. Therefore underpinning the research by a single decision theory or concept was problematic because of the danger that certain assumptions about how decisions are made would influence the research design.

Following discussions with the supervision team and the partner organisation, it was decided that the framework of treatment decision-making presented earlier in the chapter (page 69) would be utilised to inform the research and guide the study aims and objectives. This framework, which is underpinned by the legally ensconced principle of informed consent, includes at its core the concept of shared decision-making, a model of the physician-patient relationship now promoted as an ideal for the paediatric setting due to its collaborative nature and sense of shared responsibility. Because it breaks down the process of decision-making into its constituent parts, it also enabled the researcher to focus on what is contained within the ‘black-box’ of decision-making.

Although the framework has been developed in relation to the physician and adult patient dyad, it was felt that the framework offers enough flexibility to explore the triadic nature of paediatric decision-making, in addition to the varying roles that can

be assumed by the parent, young person, and professional. Thus the model can be employed “as a tool in assessing patient preferences” (Charles et al., 1999, p.658), and due to the structure of the framework, the mismatch between preferences for and participation in each phase of the process – information exchange, deliberation, and decisional control, for both the patient, and the physician, can be explored. For the current study, breaking down the analytical phase of information exchange into flow, direction, type and amount, provides the research with a clear process through which to identify the information and support needs of families for decision-making.

3.6 Chapter Summary

The rhetoric of ‘patient choice’ has driven forward new models of treatment decision-making, now reinforced by the legal requirement for health care professionals to obtain informed consent. In addition, there is a general agreement in the literature that patients should be encouraged to participate in decision-making, evident in the models of informed choice and shared decision-making discussed in this chapter. In fact, enabling individuals to have more choice and control over their care and treatment during times of illness and sickness, it is argued, is in direct response to patient demand, and leads to better outcomes for patients. Yet while the assumptions underpinning medical paternalism are now argued to be flawed, the assumptions underpinning the models of ‘choice’ are only now coming under the spotlight. Furthermore, the research concerned with patient preferences for decision-making tells a confusing, and incomplete story.

For decisions involving children, research is still in its infancy, although from an ethical and moral perspective it is argued that again, children should be involved, along with their parents, in the decision-making process. Yet how to achieve this, and whether or not it matches the preferences of children and parents, remains unclear. Although there is a growing consensus in the literature that parents and professionals increasingly recognise the benefits of working in partnership, there is

limited research about parental and child preferences for participation and control, and about the information and support needs of families for decision-making.

Research does show that for children with life-limiting conditions, decision-making is an on-going and complex process involving an array of professionals supporting the child, and a range of medical, experiential and value-based information. From the limited research concerned with decisional conflict, it can be the aspects within the process of decision-making, including the mismatch between preferences for and participation in decision-making; the failure to provide adequate information; and miscommunication and misunderstanding between patient and physician; that can lead to regret over the decision itself. For decisions involving uncertainty and complexity in which a child's quality of life and prognosis are key factors in the decision-making process, helping families weigh information of such complexity in order to make an informed decision poses an additional challenge for this process, particularly when the potential for conflict is high.

This chapter paves the way through the vast decision-making literature, focusing on the model of shared decision-making developed by Charles et al. (1999) to inform the research, and to guide the process of data collection and analysis. The next chapter focuses on the methodology employed for the research, beginning with a recap of the study aims and an overview of the methods before providing the reader with a clear account of how the research was carried out.

- CHAPTER 4 -

METHODOLOGY

This chapter provides a detailed account of the study methods, beginning with a recap of the study aims and objectives, and an outline of the methodology including a rationale for the approach taken and reference to the methodological literature. The chapter then offers further information about how the research was conducted, providing an account of the sampling strategy and criteria, an overview of the recruitment process, and details of how the data were collected, managed and analysed. Ethical considerations for the research are then presented, drawing attention to the potential impact the process of ethical review had on the study, and the ethical challenges that arose during the research process. The chapter then discusses the credibility of the research before finishing with how the researcher considered how to present the data and research findings.

4.1 Overview of Methodology

The central aims for the study are:

- To explore how decisions about technological support are made by families
- To identify the key factors involved in decisions about technological support
- To explore the interaction between families and professionals during decisions about technological support.

Two further objectives for the study are:

- To identify the information and support needs of families for decision-making
- To explore the suitability of shared decision-making as a framework within which to understand treatment decisions for children and young people with life-limiting conditions.

To examine how decisions about technological support are experienced by families and at the same time collect data about the overall process of decision-making, a qualitative approach underpinned by both interpretivism and pragmatism as recommended by Snape and Spencer (2003) was chosen for the research. Drawing on the work of Marshall and Rossman (2006), who distinguish between three main genres of qualitative research, the methodology selected focuses on the 'individual lived experience', which Marshall and Rossman (2006) point out "typically relies on an in-depth interview strategy.....to capture the deep meaning of experience in the participants' own words" (p.55).

In-depth unstructured interviews were therefore deemed to be the most appropriate and effective means to collect retrospective data about the experience of families involved in complex decisions, because parents and young people were able to provide their own narrative of the decision experience without the constraints of any pre-determined assumptions on the part of the researcher about how decisions might be made in practice. As Bowling (2009) points out, one of the key advantages to the unstructured interview is "that more complex issues can be probed, answers can be clarified and a more relaxed research atmosphere may obtain more in-depth as well as sensitive information" (p.408).

One of the key considerations in weighing up this methodology concerns the retrospective nature of the data, and Silverman (2005) points out that a participant "will document their past in a way which fits it, highlighting certain features and downplaying others" (p.8), a process he describes as a "retrospective rewriting of history" (p.8). Drawing on the notion of decision regret (Connolly and Reb, 2005), it is quite possible that subsequent events and experiences relating to the decision may influence and alter the final narrative of the original experience. Nevertheless, enabling families to highlight certain features and downplay others is the preferred choice in such an exploratory study, particularly considering the growing assumptions among policy makers and clinicians that patients wish to make decisions in certain ways, despite the lack of evidence to support these claims.

An ethnographic approach using observational methods was also considered in order to capture the interactions that take place during a decision process, and the use and exchange of information in real-time, combined with interviews to examine the process more closely through the lens of families. However, given what is known about the uncertainty surrounding decisions regarding technological support, and the overwhelming nature of taking over a child's new technology in the home, it was agreed that retrospective accounts offer more potential to capture the wider decision process, rather than the immediate impact of the decision itself.

4.2 Sampling

A purposive sampling strategy was designed for the research, defined by Bowling (2009) as "a deliberate non-random method of sampling, which aims to sample a group of people, or settings, with a particular characteristic" (p.409). The primary criterion for the sample was that the child or young person must have been diagnosed with a life-limiting condition, whether or not it was given an official name or label.

Following consultation with the recruiting organisation and supervision team, and with reference to the available literature concerned with life-limited children and young people, the decision was taken to exclude children with cancer due to the very different pathway of decision-making among this illness group, and the different circumstances under which children with cancer move from having a life-threatening to a life-limiting condition (Hinds et al., 2001).

After consulting with the supervision team and drawing on the relevant factors identified from the review of literature, a sampling strategy was designed for the research specifying key criteria for inclusion in the study in order to capture the diversity among families of children and young people with life-limiting conditions, and families with certain characteristics likely to influence the experience of making decisions about technological support.

The sampling criteria are as follows:

- ***Families who had made a decision for their child to have, or not to have a gastrostomy tube for the purpose of providing nutrition, AND / OR for their child to begin using a ventilator to assist their respiratory efforts, either continuously or intermittently, for example overnight.***

These technologies were selected as examples of interventions designed to improve and sustain the lives of children and young people with complex health care conditions, and as technologies that are often considered for children with life-limiting conditions. They were also selected for their contrasting features (see Chapter Two for further information). Gastrostomy is an invasive procedure involved with both benefits and risks, some of which relate to the surgical nature of the procedure and the risks associated with general anaesthetic, which are often heightened for many of these children and young people due to respiratory weakness.

In contrast, assisted ventilation is increasingly perceived as non-invasive because it can be effectively administered via a face or nasal mask, except for individuals who still require a tracheostomy in order to be ventilated. Both assisted ventilation and gastrostomy have in common their life-sustaining function, gastrostomy to provide adequate nutrition, and ventilation to provide adequate oxygen to the body. Both procedures, while viewed as long-term interventions, can be withdrawn.

- ***Families who decided for and against these technologies were invited to take part. This was particularly important so as to explore the experiences of those families who decided against technological support for their child.***
- ***Families who had made the decision between one and five years ago.***

The study wanted to explore the narratives of families who were able to reflect on the whole decision process. Given the existing research that shows parents are often overwhelmed by the new skills and expertise they must acquire, and may also have to deal with teething problems and minor complications

associated with their child's technological support, it was decided that families whose children had lived with (or without) technological support for more than a year following the decision would be selected for inclusion.

This decision was based on the argument that the period following initiation of technological support can be a somewhat transitional phase for families, and therefore a time during which parents may experience fluctuating feelings about whether or not they have made the right decision. It was therefore important to obtain a sample of families who were able to reflect back on this transitional phase to avoid collecting data during a period of change and emotional instability with regards the topic under investigation.

Decisions made up to five years ago would be included mainly to tackle the potential recruitment difficulties in achieving the required sample, acknowledged in the literature with regards to the area of paediatric palliative care research (Tomlinson et al., 2007), and exacerbated for this study by the ethical constraints of using only one recruitment organisation to obtain a sample. Although for some participants the recalling of specific names and times would be difficult, this particular data were not required. Moreover, the study sought to capture families' accounts of the decision process, recognising that their accounts are not in any way a re-telling of the event itself but their interpretation of that event for their child and themselves.

- ***Children and young people who were between the ages of seven and twenty-four when the decision was made, and whose cognitive abilities were developed enough for them to answer questions about the decision process.***

Including children in the study was particularly important considering their increasing legal and ethical rights in decision-making across all areas of their lives. Enabling them to share their experience of the decision-making process, regardless of their level of participation at the time, is an important step in facilitating these rights (Powell and Smith, 2009). In fact, as academics and policy makers increasingly recognise the importance of obtaining the views and

experiences of children, their 'voices' are becoming the focus of a slowly growing body of research in social sciences across the disciplines. Furthermore, studies that have harnessed the world of children as lived and viewed from their own perspective, have reported the data to be of a depth and richness beyond expectations (Rabiee et al., 2005; Earle et al., 2006; Sarvey, 2008).

It is suggested in the literature that children from the age of around seven are able to participate in decisions that affect their lives (Friedman Ross, 1996). The level of participation will of course depend on their capacity for decision-making, normally assessed by taking into consideration their developmental age, maturity, and decision-making experience. It will also depend on the views of their parents and the professionals involved in their care about how much participation children should be afforded.

At the upper age limit, the transition young people make into adulthood is a complex one, and while this study has included decisions for young people up to the age of twenty-four, it is accepted that many young people regard themselves as an adult sometime before this. The upper age limit also acknowledges that young people with life-limiting conditions are unique, their life-course following a trajectory quite distinct to other children due to their increasing dependence on their parents and/or adult carers.

It was decided that children and young people with moderate to severe learning disabilities would be excluded from taking part in the study, although parents of these children would still be invited. This decision was based on the purpose of interviews, which would require children to both recall an event that had taken place in the past, and to have an understanding of their condition, and their care and treatment (Greig et al., 2007). The potential distress of including children and young people with a limited awareness and understanding of these issues, and the limited data to be gained from them, outweighed any potential benefits to including them in the study.

- ***A sub-sample of bereaved parents***

A small number of bereaved families were invited to take part in the study. Drawing on the concepts of decision regret and cognitive dissonance, which may be experienced differently once a child has died (Beresford and Sloper, 2008), it was hoped that bereaved families would offer a more reflective account of the decision process which families who are still experiencing the outcome of the decision they made with regards to technological support may not.

In addition to the above criteria, efforts were made to include the following with the objective of capturing the range and diversity of views and experiences on the research topic:

- Children with a range of diagnoses and prognoses
- Families who had access to varying sources of support at the time of making a decision (e.g. a hospice, specialist nursing team)
- Families of varying compositions with regards to marital status, ethnicity, employment, socio-economic background, presence of other children in the family.

Because of the exploratory nature of the research and the level of depth required from the data in order to meet the study aims, an initial sample size of twenty families was considered sufficient in order to capture the complexity and range of decision-making experiences. The final sample size would be guided by the recruitment process and in monitoring the level of data saturation achieved from the sample (Marshall, 1996).

4.3 Recruitment

The original study design included three separate research sites for identifying and recruiting the sample, two children's hospices and an NHS site, so as to include families not using a children's hospice, and in order that the sample could be achieved. However, following the process of ethical review only the collaborating

organisation could be utilised to recruit families due to the additional resources required to be in place to support both the researcher and the research participants should they become distressed as a result of taking part (see Section 4.7 for more details). Therefore the entire sample was recruited from users of the children's hospice, which provides support to children and young people with life-limiting conditions and their families across the Yorkshire and Humber region.

The sampling strategy was used to guide the recruitment process and to ensure the sample was obtained in a reliable and robust manner. Nevertheless, it was argued by the collaborating organisation that due to the exploratory nature of the study and the difficulties in recruiting families living under such difficult circumstances to participate in research, a problem also acknowledged by Tomlinson et al. (2007), that the criteria should be employed flexibly, and families considered for inclusion on an individual basis. In order to achieve the sample, an agreement was made to approach recruitment in this way, but to have regular meetings with the children's hospice manager about the progress being made in achieving the sample.

Because the research relied on the recruiting organisation to apply the sample criteria to the selection process, the regular meetings with the hospice manager were also used to ensure that there was no potential bias in the selection process. The only additional criteria used to assess a family's suitability for inclusion in the research relates to how recently they had been invited to take part in other research (the hospice has a policy of not inviting families to take part in new research if they have taken part in other research within the last three years as a result of being approached by the children's hospice), and on ethical grounds (for example if it was felt that the family were under additional stress during the recruitment phase of the project, such as the child entering the end of life stage).

In order to explore the decision-making process from the perspective of those involved in decisions about technological support, both parents where possible, or a parent and other family member identified as important in the decision about technological support, along with children and young people where appropriate,

were invited to take part in an in-depth interview with the researcher. Invitation packs were sent to potential participants containing a covering letter from the recruiting organisation (see Appendix C); an information sheet (versions were created for parents of children under 16, parents of young people aged 16 and over, bereaved parents, young people age 16 and over, older children (eleven to fifteen), and younger children (seven to ten) [see Appendix A]); and a response form (see Appendix B). All versions of the information sheet were piloted with parents / young people, and modified accordingly.

Potential participants were asked to complete the response form enclosed in the pack and return it to the researcher. This method of recruitment was utilised to ensure that participants voluntarily, without persuasion from a third party, put themselves forward to take part in the study having been fully informed about what this would involve. To ensure participants had time to make an informed choice, they were given two weeks after receiving the information leaflet to consider fully the implications of taking part, following which they were sent a reminder letter.

Families who expressed an interest in taking part in the study were contacted by the researcher, during which further information about the study was offered, and an opportunity for families to ask questions and raise any concerns they had about taking part was provided. After checking again for consent, the researcher arranged to visit the participants to conduct the interview either at their homes, or another suitable place of their choosing that offered relative privacy.

4.4 Achieving the Sample

The recruitment of families to the study was slow and arduous. A number of meetings were required during this phase of the research to review the sample criteria and discuss how to recruit more families. Because response rates were extremely low, which was problematic due to the criteria for the study and the limited number of families from which to select a potential sample, the invitation packs were revised to include a short letter from the recruiting organisation and a

summarised information sheet about the project with the intention of providing a full information leaflet once a response form was returned. It was hoped this would encourage families to take the time to look over the shortened information.

Ultimately however, the sample criteria had to be relaxed further in order to obtain the sample required. After discussing the options with the supervision team, which were to either extend the time span for the decision to ten years, or to introduce another technology (spinal rods) to the study, it was decided, due to the practical constraints of the latter which included going back to the ethics committee and recruiting further bereaved parents when this sample had already been achieved, that families who had made a decision within the last ten years would be included in the sample.

Interestingly, bereaved parents proved the least difficult to recruit, and of the initial sixteen bereaved parents who were invited to take part in the study, seven agreed and were interviewed, giving a response rate of 44%. This compares to a response rate of 21% for other families (thirty-four families were invited to take part in order to obtain seven families).

However, the stage at which fourteen families had taken part in the research, it became evident that recruiting children and young people was proving to be impossible, and of the fourteen families recruited over a period of several months, seven were bereaved parents, and five had children with no form of communication. Of the remaining two, the parents had no wish to involve their child in the study.

Following a meeting with the collaborating partner to discuss the reasons for this it was decided that only young people aged 18 and over (in other words those who in the view of the health care system were independent decision makers or adults) would be invited to take part. It was also decided that the approach would change for this part of the sample in that, in order to respect the autonomy and decision-making capacity of young adults, parents would only be invited to take part in the research where the young person agreed to take part and following their interview

gave the researcher permission to contact their parents about the study. This method facilitated the recruitment of six young people (14 were invited giving a response rate of 43%), three of whom agreed for their parents to be approached by the researcher.

4.5 Data Collection

Nineteen families were recruited to the study. In total, nine bereaved parents; ten parents of children and young people with no or very limited decision-making capacity; six parents of young people with capacity for decision-making; and five young people (aged between 22 and 32) took part in the research, each of whom had been involved in a decision about gastrostomy or ventilation and in some cases both. An overview of the sample is provided in Table 2 below.

Table 2 – Sample Overview

Details of the Study Sample	
<ul style="list-style-type: none"> ▪ Six families of children with no or very limited decision-making capacity, either due to age or the nature of their condition. 2 mothers, 4 couples (mother and father) 	
<ul style="list-style-type: none"> ▪ Seven bereaved families. 3 mothers, 3 couples (mother and father), 1 father Among the bereaved families, four children had lived with no or very limited cognitive abilities and therefore were not involved in the decision about technological support. Three young people had reached the age of having either developing or full decision-making capacity. 	
<ul style="list-style-type: none"> ▪ Six families of young people with either developing or full decision-making capacity. 3 mothers, 2 couples (mother and father), 5 young people For the only young person below the age of 16, the parents decided to take part but did not wish for their son to take part in the study. Two young people did not wish for their parents to take part. Three young people were interviewed and agreed for their parents to take part. 	

The data were collected through in-depth face to face interviews, which were recorded using a digital audio recorder. A single interview was carried out for parents, and where both parents took part they were interviewed together. Separate interviews were conducted with young people.

Topic guides were developed for parents and young people (see Appendix E), drawing on the model of treatment decision-making underpinning the study to include the separate phases in the decision process and the different levels of participation. The interviews were mainly unstructured to enable participants to provide their own account of the decision process, and the topic were employed as a tool to guide discussions towards key topics where necessary and to ensure that questions specific to the process of decision-making were asked consistently across the sample.

To acknowledge the development of topic guides as a key stage in the research process is important, and as Arthur and Nazroo (2003) point out “a well-designed topic guide will provide flexible direction to fieldwork process and essential documentation of a central aspect of the research. A poorly designed topic guide at best will be confusing and at worst will restrict the exploratory and reflective nature of qualitative research” (p.115). Therefore considerable time was taken to develop the topic guides to ensure the data collected would have both depth and richness, and provide consistency across data collection.

Initially the guides were built from the literature review, and then modified as the study aims and objectives were finalised. Rather than a designing question-based guide, the guides were divided into key topics, each broken down into issues that needed covering in the interviews. This approach is recommended by Arthur and Nazroo (2003) who suggest that topic guides structured like questionnaires lack the flexibility required to obtain ‘in-depth’ data and to pursue avenues of interest not contained within the topic guide, yet relevant to the study.

Because the study sought to collect descriptive and factual data relating to the aims of the study, as well as exploring the decision process, it was important that the

topic guides struck the right balance between the structure required to collect data about preferences for participation in decision-making and needs for information and support, and the flexibility required to enable participants to openly share their experience and perspectives of the decision-making process, which has been found to enhance memory recall and accuracy (Krähenbühl and Blades, 2006).

Participants were informed of the main topics to be discussed at the beginning of the interview in order to enable participants to discuss the topics of relevance to the project and at the same time integrate issues they felt to be important into their own narrative. For parents the topic guide was only employed when necessary, in the main to pursue those topics participants either did not talk about freely or where further depth was required.

Two pilot interviews were conducted to test the usability of the topic guides, one with a ventilated adult male with a diagnosed muscular degenerative condition, and one with a parent of a boy who is fed by gastrostomy and has other complex health care needs. The data were examined for richness and depth, and feedback was provided by the participants regarding the interview topics. The topic guides were then modified further, with additional probes, or 'branches' inserted to ensure the data offered more depth than was achieved during the pilot interviews.

In order to provide a safe and comfortable environment for family participants to share their experience, which for some was emotionally challenging, the researcher adopted an informal approach with families, and paid attention to the impression her clothing and general appearance could make on the power imbalance between the researcher and the researched. During interviews, active listening skills were employed, expressing interest and attention, and adopting an open and friendly body language. To achieve this, topic guides were committed to memory to avoid unnecessary distractions, unnatural pauses, and obvious changes in body language.

The approach taken by the researcher during young people's interviews varied depending on their age, confidence and understanding. The techniques used during interviews were also informed by existing research about how to effectively

interview children and young people (Instone, 2002; Krähenbühl and Blades, 2006; Almerigogna et al., 2008), and a training course concerned with how to effectively consult with disabled children and young people attended as part of the research process. Compared to parent interviews, more time was taken to build rapport with young people, asking questions about school, hobbies and other interests during the early stages of the interview. The researcher also made efforts to position herself at an angle to the young person, rather than directly in front of them to help make them feel at ease and to minimise the formality of the interview situation. The topic guides were employed flexibly to encourage responses, and where young people were particularly shy, the first part of the interview focused on questions that young people could answer easily, without too much thought, before moving on to more open-ended and reflective questions.

4.6 Data Analysis

It is important to note that while the stages of data collection and analysis are presented separately here for the reader, they are not mutually exclusive, and instead considered in combination as an interactional process, each influencing the other from the outset (Pope et al., 2000). The process of analysis began as the first data were collected, through the process of familiarisation or immersion, and also researcher reflection. As questions were asked of the data, and early themes and categories emerged, new avenues of inquiry and areas requiring further probing were utilised. Each subsequent interview therefore followed a unique trajectory, shaped in some way by the collection and analysis conducted to date. In order to facilitate this process, reflective field notes were made following each interview which assisted the researcher in the early stages of analysis and also when reflecting on the findings to look at the ways in which data collection may have influenced data analysis, and vice versa.

4.6.1 Using Framework to manage the data

The Framework approach to analysing qualitative data was adopted to manage and code the data for further analysis, and also informed the analytical process. This

method of analysis was first published by Ritchie and Spencer in 1994, and has been modified over the years as a method of analysis particularly suited to applied or policy relevant research (Ritchie et al., 2003; Spencer et al., 2003). As well as offering the researcher an auditable process for analysing qualitative data, it enables the researcher to simultaneously conduct analyses in a deductive and inductive manner, drawing on the data for emergent themes and categories and also analysing the data with regards to the aims and objectives determined from the outset (Pope et al., 2000; Ritchie et al., 2003).

For Ritchie and Spencer (1994), “qualitative data analysis is essentially about detection, and the tasks of defining, categorizing, theorizing, explaining, exploring and mapping are fundamental to the analyst’s role” (p.176). The method of Framework offers a process of analysis that integrates these tasks, and there are five recognised stages of Framework – familiarisation; identifying a thematic framework; indexing; charting; mapping and interpretation (Ritchie and Spencer, 1994).

The first four stages were applied to the research data, and although an integral part of the analytical process, these stages of labelling, sorting and synthesising are often referred to as data management, and distinguished from the final stage of mapping and interpretation which for Ritchie and Spencer (1994) is the point at which “the serious and systematic process of detection begins” (p.186).

Nevertheless, Ritchie et al. (2003) note that “through all the stages of data management, “meaning” is being attributed to the original material” (p.237).

The process of familiarisation began with transcription, as the researcher listened to and transcribed the raw data. Transcripts were then read and re-read to gain an insight into the depth and richness of the data, and to identify key ideas and recurrent themes (Ritchie and Spencer, 1994, p.179). The information gathered during this stage, along with a priori issues informed by the research aims, were used to identify the beginnings of a thematic framework, or detailed index of all the key issues, themes and categories in the data. In order to create a manageable

framework, the index was hierarchically structured by main and sub-themes. Each item in the framework was given a descriptive label and also assigned a number for the purpose of indexing (see Appendix H – Thematic Framework for Data Analysis).

The nature of the data, as unstructured narratives about particular events loosely framed around the concept of shared decision-making that underpins the research, allowed for an analytical approach involving both deductive and inductive methods. The framework therefore includes a priori themes derived from both the framework of shared decision-making and the aims of the project, as well as themes identified within the data during the analytical process. During the data familiarisation and coding process it became clear that both pre-determined and emerging themes related to the decision itself and the process of decision-making around it. It also emerged at this point that certain features of decision-making for children and young people with life-limiting conditions were also relevant to decisions.

The thematic framework was then applied to the data, in other words the data were indexed. Each segment of data was indexed using the assigned numbers, some several times in cases where more than one theme or issue was of relevance, referred to as multiple indexing (Ritchie and Spencer, 1994). Throughout the early stages of indexing the thematic framework was revised to reflect more accurately data coverage and diversity. At each addition and revision the data already indexed were re-visited with regards to the modifications made. Annotations were made to those indexed segments requiring some consideration to the interpretation or meaning for later reflection.

The data were then sorted by theme or concept, a process referred to as 'charting', which for Ritchie and Spencer (1994) enables the researcher to "build up a picture of the data as a whole, by considering the range of attitudes and experience of each issue or theme" (p.182). The process of charting involved creating a chart, in Microsoft Excel, for each key theme within which indexed segments of data were placed for analysis. These charts became the focus for further analysis, in other

words the process of interpreting and explaining the findings and data, details of which are provided in the following section.

4.6.2 The analytical process

The analytical process followed the approach described by Ritchie et al. (2003), who employ the concept of an analytic hierarchy, “a series of ‘viewing’ platforms, each of which involves different analytical tasks, enabling the researcher to gain an overview and make sense of the data” (Spencer et al., 2003, p.213). The analytic hierarchy not only depicts the stages involved in this type of qualitative data analysis but enables the researcher to move up and down the hierarchy in order to re-visit the data where necessary, and portrays the way in which the process of analysis moves towards higher levels of abstraction in order to develop explanations and consider wider policy and theoretical applications.

Mapping and interpretation is sub-divided by Ritchie et al. (2003) into ‘descriptive accounts’ and ‘explanatory accounts’, each a process on the analytic hierarchy, their difference in the level of abstraction desired. Descriptive accounts tend to remain close to the data, and the process involves defining concepts, refining categories, and establishing typologies. It involves three key steps – detection, categorisation, and classification – and aims to “unpack the content and nature of a particular phenomenon or theme” (Ritchie et al., 2003, p.237). The findings presented in Chapters Seven, Eight and Nine represent the result of this process.

‘Explanatory accounts’ are developed later through a process of detecting patterns and associations, providing explanations, and seeking wider applications. It is at this higher level of abstraction that consideration is given to the study’s wider applicability, at a theoretical or policy level (Ritchie et al., 2003), and it is through this process of analysis that a new model of treatment decision-making for children and young people with life-limiting conditions was developed. Whilst clear links can be seen between the model and the thematic framework into which the data were charted (see Appendix H), the thematic framework alone is insufficient as a tool through which to understand the complexity of decisions about technological

support because it represents only the indexing and charting, and to some degree the descriptive analysis of the data.

It is in the investment of additional analytical work in looking for patterns and associations, and seeking wider applications that has enabled the researcher to offer an applied model through which to understand the complexity of decision-making for children and young people with life-limiting conditions. This stage of analysis followed the process described by Ritchie et al. (2003). However in doing so other methods of analysis were consulted, and the influential work of Glaser and Strauss (1967) was particularly helpful because in developing the model and considering the relationship between the different elements of decision-making, the constant comparative method was applied to ensure the model represented the data being analysed. The approach described by Marshall and Rossman (2006) was also useful, and guided the researcher to continually look for alternative explanations for the patterns and associations found during the analytical process.

4.7 Ethical Considerations

Due to the sensitive nature of the project, both in respect to the research population and the topic under investigation, the study was underpinned by an ethics of care. In particular, the process of informed consent; the nature of confidentiality and anonymity; the sensitivity of the project; and the participation of children and bereaved parents were identified as key areas to address during the project design phase.

4.7.1 Informed consent

Informed consent was viewed as an on-going process throughout the recruitment and participation phases in the study, beginning with process of recruitment detailed in Section 4.3 which was in part designed to ensure potential participants were able to make an informed and voluntary choice free from coercion, about whether or not to take part in the research. Prior to interview, participants were again asked if they were happy to participate and given another opportunity to ask

further questions. They were then asked to sign a consent form (see Appendix D). If a participant was not happy to sign a consent form, or was unable to because of a physical impairment, then verbal consent was recorded at the beginning of the interview.

This process of checking and re-checking consent, and providing several opportunities for participants to ask questions about the project, ensures that participants are able to make an informed choice about taking part in the project. This process was to continue throughout the interview by checking for verbal and non-verbal signs of distress or reluctance to participate, and by ensuring that participants were given time to talk about their participation following the interview if they wished.

Separate consent was obtained for recording interviews (audio) and transcribing the data collected during family interviews. Participants were informed that they could withdraw from the study at any time during and following participation but that at a later stage their data may already be analysed and published. At this stage though, their transcripts would be destroyed.

4.7.2 Confidentiality and anonymity

A potential sample was approached in the first instance by the recruiting organisation, who used confidential data about the families they have supported to ensure families met the criteria for the study. The researcher did not have access to this data and was only able to contact potential participants following contact initiated by them. Participants were assured that everything they told the researcher would be held in confidence, and that any quotations included in published reports or papers would be unidentifiable. All participants were informed immediately before their interview that confidentiality would only be broken if they disclosed information that identified them, or some-one else, as being at risk of significant harm.

Williamson et al. (2005) point out that consideration of how to phrase this breach of confidentiality to children and young people is important because failing to

adequately address the conflict between confidentiality and child protection threatens the process of informed consent. They argue that a child's perception of harm may be very different to that of an adult, and yet talking explicitly about child abuse and protection is inappropriate. Children and young people under the age of sixteen were therefore to be informed, in age appropriate language and terminology, that firstly their right to confidentiality is limited by the researcher's moral and ethical code to protect the safety of child participants, and secondly that should a child or young person reveal information suggesting that they, or another minor, are at risk of harm, this information would have to be discussed with an appropriate adult. However, the decision about which adult to discuss the information with would be shared with the young person.

With regards to protecting the identification of participants, once the audio data had been transcribed the digital data were destroyed. Transcripts contained only an identifier code, assigned to participants to ensure that should they subsequently decide to withdraw from the study, their transcript could be identified by the researcher and destroyed. Transcripts contained no reference to any named person, service, or place. The only documentation containing a participant's identity, which included details of assigned codes on transcripts and other documentation, and the signed consent forms, were stored separately to the data in a locked filing cabinet. Only the researcher has access to this information.

4.7.3 Sensitivity of the project

Tomlinson et al. (2007), exploring the challenges to participation in paediatric palliative care research, identify four ethical concerns regarding sensitivity – the burden of participation on families; the potential vulnerability of families; the possibility that parents are yet to accept their child's impending death; and the possibility that the child is not aware their life is limited (p.436). Three strategies were established to address these challenges – ensuring the process of informed consent enabled families to make voluntary, fully informed decisions about participation, with the option of withdrawing at any time; interviewing parents

before children therefore enabling the researcher to structure the interview and questions for children around the unique circumstances of each family; and ensuring that adequate support was available for families who may experience distress as a result of reflecting on a particularly emotive decision.

The final strategy of providing support involved closely monitoring verbal and non-verbal signs of distress during the process of consent, and throughout and following the interview to ensure that those families in need of support were offered it. As well as written information regarding available information and support (see Appendix G), families requiring immediate support were able to contact the consultant clinical psychologist at the collaborating hospice for assistance. If they preferred, the researcher would arrange this for them. Similar support was also made available to the researcher, due to the emotional aspects of the topic under investigation, a recommendation set out by Mitchell and Irvine (2008) who highlight “the need to think more carefully and proactively about researcher well-being and the need to focus on issues of emotional risk” (p.40).

4.7.4 Participation of bereaved parents

Additional concerns about the participation of bereaved families were raised, due to their particular vulnerability. However, bearing in mind Tomlinson et al.’s (2007) observation that there is “a tendency for overly paternalistic protectionism that can limit the progress of palliative care” (p.436), it was decided that, on ethical grounds, the measures in place to minimise the potential distress to participants were sufficient for bereaved families. Furthermore, it was felt that excluding bereaved families from the study simply due to their bereavement was unacceptable, and it was therefore more ethically appropriate to offer them an opportunity to talk about their experience especially considering that participation was based on the principle of voluntary and informed consent.

In fact, a recent study exploring the perceptions among bereaved parents regarding their participation in research found that, while it could be painful, it was also a positive experience that enabled parents to tell their story in an environment of

respect and confidentiality (Dyregrov, 2004). Hynson et al. (2006) agree that “the research process itself can be seen to offer potential benefit to bereaved parents when undertaken with care and sensitivity” (p.811). They add that in-depth interviews as a method to collect data was perceived as particularly suitable for bereaved parents, who talked about the importance of being able to have control over the pace and content of discussions. However, appropriate training for interviewers was argued to be essential. Therefore, as well as employing in-depth interviews as a method for collecting data, the researcher underwent additional training on both bereavement and in-depth interviewing.

Hynson et al. (2006) also emphasised the importance of timing the approach of participation for bereaved parents, which was set at 6 months following bereavement for their study. After taking advice from the collaborating hospice, who provide a bereavement service to the families they support, it was decided that only parents whose child had died at least 12 months prior to recruitment would be invited to participate in the study. The recruiting organisation would, in addition, consider the suitability of bereaved parents on a one-by-one basis to minimise any potential distress to participants.

4.7.5 Participation of children and young people

The shift towards including children in qualitative research has led to an acknowledgement of the unique status of children as research participants, and the process of informed consent required (Friedman Ross, 1998; Williamson et al., 2005; Gibson and Twycross, 2007; Coyne, 2009). In respecting the legal status of young people regarding consent, it was decided that for this study young people aged sixteen and over would be sent separate invitation packs to their parents, even though many of them still lived at home. Because their parents were also being invited, both the young person and parents were informed of the other’s invitation to take part in the research. With regards to consent, parents and young people were treated as separate participants, and consent obtained on this basis.

Deciding on an approach to invite children and young people under the age of sixteen was more difficult because although legally it is still the case that parents must consent on their behalf, the research wished to acknowledge the growing evidence that children below the age of sixteen often have capacity for decision-making. Following consultation with the collaborating partner, a family approach was adopted, recognising the importance of enabling children and young people to be involved in the decision to participate, but also acknowledging both the authority of parents in decision-making, and the level of expertise and knowledge they have regarding their child.

Therefore parents would be sent an invitation pack inviting them and their child to take part in the study. Parents were asked to provide their child with the enclosed invitation if they believed it was appropriate for them to be involved. As well as consent from parents regarding their child's participation, assent would also be required from children and young people. In order to gain assent, age-appropriate information sheets and consent forms would be provided so they could make an informed choice regarding participation (see Appendix A and D).

An essential component of any ethically driven research involving children is to ensure their participation yields useful data, partly to make sure that it does not become merely a token gesture in the acknowledgement of children's increasing rights, but also to include their views as a central component of the research process. It was important then, that children viewed their involvement as a positive experience by engaging them in age-appropriate activities that enabled them to share their views with the researcher (Rabiee et al., 2005; Powell and Smith, 2009). As well as using appropriate methods to obtain useful data, the researcher took up additional training on consulting and communicating with disabled children.

Engaging children effectively in the research was also an essential step towards addressing the power imbalance between adult researcher and child participant (Christensen and James, 2008; Powell and Smith, 2009), and minimizing the potential distress of participation given the vulnerability of children with complex

health care needs. Prior to interviewing children, it was also important to be aware of the impact of their condition on the interview process to ensure children were not interviewed for too long, and that appropriate breaks were provided. For children who tired quickly, families were informed that the researcher could carry out the interview over more than one visit. Finally, verbal and non-verbal cues of dissent were monitored very closely in children's interviews, again to minimise potential distress, and in awareness of children's limited power in the research process.

4.7.6 *Assessing the capacity of young people to consent*

The approach to obtaining the consent of young people aged sixteen and over was based on the principle underpinning the Mental Capacity Act 2005 that individuals are assumed to have capacity for decision-making (DCA, 2007). However, it was also acknowledged that due to the research population, some of the young people being invited would have cognitive or communication impairments that would call into question this assumption, and that they may lack the capacity to make a decision about taking part in the research. The problem of assessing capacity was discussed at length with the recruiting organisation to ensure that young people were encouraged to take part but that the approach to consent was appropriate for individual families.

Having consulted with the recruiting organisation and referred to the Mental Capacity Act 2005 guidance (DCA, 2007), it was decided that a range of different packs would be made available to the recruiting organisation to cover the various consent / assent scenarios that might arise during the recruitment process. In addition, the individuals responsible for the recruitment process were selected because of their knowledge of families using the hospice, and agreed to ensure that the capacity of young people was assessed to determine which pack to send. Where it was felt that young people aged sixteen or over had learning disabilities or other impairments that would call into question their ability to consent in their own right,

packs were sent to parents, with parents being asked to provide a pack to their adult child after considering whether it was appropriate.

4.7.7 Obtaining ethical approval

The study was approved by the Humanities and Social Science Ethics Committee at the University of York following minor amendments to participant documentation in May 2008. An application was then submitted to the National Research Ethics Service (NRES), established by the NHS to protect the rights and wellbeing of research participants and promote ethical practice. This process is required for all research involving NHS staff, patients, users, and also individuals identified for research purposes because of their status as relatives or carers.

The initial application, following review, was rejected in June 2008 (see Appendix F for copy of letter) on the following grounds:

- Lack of training and experience of the researcher due to the sensitive research population and topic under investigation. Furthermore, the rejection stated that “a person with clinical experience in supporting families in these circumstances should either conduct the interviews or be in attendance at the interviews to support the Chief Investigator”. This concern had serious implications for the researcher as a postgraduate student who had the necessary ‘research’ training but no clinical experience or qualifications.
- Lack of immediate support following interviews for both the researcher and families. The procedures in place were felt to be inadequate.
- The Committee felt that the consent process regarding families in which conflict may arise about participating in the project was unclear. For example, a family in which parents wished to participate but their child did not want either themselves or their parents to participate, could create intra-family conflict around participation. The researcher intended to deal with such situations as they arose, working with the recruiting organisation

in order to resolve any potential conflict around participation. The Committee felt that in this situation families should be excluded from the project at the outset.

- As the study proposed to include young adults who may lack decision-making capacity, additional procedures for assessing capacity under the Mental Capacity Act 2005 were set forth. The committee felt that the proposed arrangements for assessing capacity were inadequate.

Under the provisions laid down by the National Research Ethics Service, the researcher was entitled to either appeal against the decision in which case an identical application would be reviewed by a different Committee, or to submit a new application which would contain within it details of the rejection and an explanation of the changes made to address the issues raised. Following a discussion with the supervision team, the decision was made to submit a strengthened application which addressed the concerns raised by the first Committee.

In particular, a solid and robust argument defending the researcher's skills and training as appropriate to conduct the study was built into the application. A set of criterion for informed consent was established in order to deal with potential intra-familial conflict around participation. Clear procedures were developed to ensure adequate support could be provided for both participants and the researcher, including debrief for the researcher with a member of the supervisory team who has specific experience in providing psycho-social support.

With regards to sensitivity, it was decided that the recruitment gateways would be limited to the collaborating partner, a children's hospice, due to their ability to advise on suitable families for participation and to provide adequate support throughout the study. This revised approach was in response to the objections raised during ethical review about the possibility that families, through the process of talking about decisions that may have involved controversy and conflict, may become distressed and require independent support following participation.

It was also decided, following a review of the Mental Capacity Act 2005 (DCA, 2007; Richards and Mughal, 2009), that adults who lacked capacity would not be invited to participate in the study. This was partly due to the limited resources available to the researcher to assess capacity to participate but more importantly, it was agreed that the additional information to be gleaned from this group of young adults, who were either not involved in decisions about technological support due to their incapacity, or whose capacity had since lessened due to the progressive nature of their condition, would not be sufficient to justify their inclusion in the project when compared to the risk of distress caused through participation.

A strengthened and revised application was submitted to the National Research Ethics Service (NRES) in July 2008. Following review in September 2008, the application was given a provisional opinion subject to addressing a number of concerns (see Appendix F for copy of letter). Some of the concerns were straight forward requiring information sheets and consent forms to be amended, and additional information regarding funding to be provided. However, the central concern raised during the first review about the sensitivity of the project and the potential for unresolved conflict associated with decisions about technological support to re-surface, was raised again. This time, though, it related to the remote possibility that families, through the process of reflecting on their experience, may consider making a complaint against a professional or service provider regarding the care or treatment of their child.

The Committee recommended that all potential families should be made aware that if an official complaint regarding their child's care and treatment resulted in a court hearing, the transcript of the research interview could be requested. It was recommended that this information should be included in participant information sheets. However, the researcher argued that this possibility was very remote, that only relevant information would ever be requested, and that transcripts would be anonymised and therefore unlikely to be submitted as evidence. The Committee agreed that only families who agreed to participate would be informed, and that this could happen during the process of consent as long as a statement was

included on the consent form to verify that participants had been provided with this information.

The initial concern regarding potential family conflict over participation was again raised. The guidelines for inclusion were still felt to be inadequate because they failed to address the situation in which a parent agrees to participate in the study but their child not only refuses to participate, but is unhappy about their parents' participation. It was agreed that response forms for families with children under the age of sixteen would include a statement about their child's feelings regarding their own participation. However, it was argued that for young people age sixteen and over, it was more important to respect their status as fully consenting adults, ethically and legally. Final approval from NRES was received in November 2008 (see Appendix F for letter of approval).

4.7.8 The impact of ethical review on the research

It was useful to reflect on the process of ethical review and how it may have shaped the research process, and the researcher, particularly during data collection. In particular, three of the concerns raised during ethical review had the potential to compromise the process of data generation by allowing the potential for bias to occur. The first relates to the requirement for the researcher to discuss breaches of confidentiality with parents in relation to any court proceedings regarding the care and treatment of their child immediately prior to interview. This requirement may well have biased the process of data collection, bringing into focus for participants the negative aspects of their child's care and treatment, or alternatively causing families to avoid these issues during the interview. In addressing this the research used a carefully worded but light hearted script, designed by the research team and piloted with members of an existing parent consultation group established in the Social Policy Research Unit.

Secondly, the concern regarding the potential for intra-familial conflict to arise as a result of enabling different family members to participate in the study led to the potential exclusion of certain families from the research. In particular, it excluded

those cases in which a parent wished to participate but their child was not happy for them to do so, as well as those families in which a child wished to participate but their parents were not willing to give their consent. The data collected from these families may well have offered a different perspective to those families whose members were in agreement, and may well have provided an insight into those situations where children and their parents have conflicting views about participation in decision-making.

Thirdly, due to the sensitive nature of the project it was decided that children and young people under the age of sixteen would be interviewed after their parents. Adopting a family approach enabled the researcher to understand the extent of a child's knowledge regarding their condition and the decision process before interviewing them. However, in doing so the depth and richness of data regarding young people's own preferences for participation in decision-making was compromised, due to the limited extent to which the researcher could probe young people whose parents expressed strong beliefs about minimizing their child's role in decision-making. Nevertheless, the ethical duty to minimize harm outweighed the need to generate data that could alter the dynamics of the family unit, which for families of children with a life-limited illness can already be under strain.

Although the ethical review caused delays and led to some changes regarding the sample and recruitment of families, it was also an advantage for the research because it forced the researcher, in consultation with the supervision team, to ensure the methods and approach to the study were robust, and to regularly monitor the progress of the study and the process of data generation. Thanks to the on-going support from the recruiting organisation, the sample achieved was purposive and generated data of great depth and range.

4.7.9 Ethical challenges during the research

Two ethical challenges arose during the course of the research. One concerns a bereaved parent who became extremely distressed during the interview. The other is about the inclusion of a young adult who had been assessed to have capacity to

consent by the recruiting organisation, but who the researcher felt should not have been approached for inclusion in the study. The strategies developed during the process of ethical review enabled the researcher to respond swiftly and purposely to these challenges, and to receive feedback from a member of the supervisory team at the earliest opportunity.

For the matter concerning the capacity of one of the young people, who was staying at the children's hospice during participation, the researcher consulted a member of the supervisory team prior to interviewing the young person. This individual was able to speak to the young person involved to ensure they had an understanding of the project and what taking part involved. In addition, the young person was asked if he would like a known adult present during the interview. This was recommended because of the communication difficulties this young person has, and the anxiety he has expressed about new people not being able to understand him. This offer was welcomed by the young person, who was accompanied during the interview by a member of the care team already known to him.

In dealing with the distress experienced by one of the bereaved parents taking part, the researcher was able to draw on both the training received around in-depth interviewing and bereavement, and on the literature consulted as part of the ethical review around doing research with vulnerable groups. Having resources for support to hand was also useful, and the researcher spent time with the parent following the interview to discuss any needs they might have for support. The researcher then debriefed with a member of the supervisory team to discuss her concerns about the participant's well-being and mental health. Drawing on the different stages of grief and how they related to this individual parent enabled the researcher to deal with the inner conflict arising from this difficult interview.

4.8 Credibility of the Research

One of the important developments to come out of the exhausted methodological debate which polarised quantitative and qualitative research for many years

(Bryman, 1984; Rich and Ginsburg, 1999; Payne et al., 2004), is the increasing popularity of methodological pluralism and the ascendancy of qualitative methods “to a level of parallel respectability” (Patton, 2002, p.265). The acknowledgement that qualitative research is a robust method of enquiry is in part due to the on-going pursuit for appropriate criteria by which to measure the quality of qualitative research, and there is a growing consensus that the conceptualisations of ‘validity’, ‘reliability’, and ‘generalisation’ associated with quantitative methodologies must be challenged and reconfigured if they are to have any purpose for evaluating the credibility of research using qualitative methods (Silverman, 2005).

To ensure the study was of high quality, the research was informed by the strategies described by Lewis and Ritchie (2003), and by Silverman (2005; 2006), referring in particular to the criteria developed by Silverman (2006) for evaluating the credibility of qualitative research (p.276). Adopting a rigorous approach, both conceptually and methodologically, was a key goal for the research, and the use of a purposive sample, the attention to potential biases in the data, the use of data saturation in determining sample size, the auditable stages of analysis and the level of abstraction achieved from the analytical process, and the consideration of how to present the findings provide evidence of the research quality and credibility.

4.9 User Involvement and Project Consultation

Regular consultation took place with the partner organisation throughout the research. Staff and users of the children’s hospice were informed about the research and utilised to obtain feedback regarding recruitment materials and interview topic guides. The project was discussed with an existing Consultation Group of parents of disabled children set up by the Social Policy Research Unit (SPRU) at the University of York. This Group also provided feedback about the recruitment materials and interview topic guides. The Children and Families Research Team at SPRU provided useful input throughout the research, and an existing research committee comprising practitioners and academics working in

paediatric palliative care across the region were also involved, particularly during the design phase and in helping to determine the sampling strategy.

4.10 Presenting the Findings

Over recent years, the wider impact of research has gained attention among policy makers and within the research sector itself (Research Councils UK, 2011). In acknowledgement of this trend, and in considering the applied nature of the project, attention was given to how the findings of this research should be presented. Focusing on the intended target audience of healthcare professionals and service providers, and on the exploratory nature of the research, particular consideration was given to the use of pseudonyms, the inclusion of direct quotations, the tense in which to present the data and the structure of the findings.

With regards to the structure, the model developed from the research is presented at the earliest possible stage. Details of the model are contained in the chapters that follow the model, drawing on the key themes in the model and using the data to illustrate. A number of case studies have also been provided at the beginning of the findings. This approach has been adopted due to the limited knowledge and understanding among generic health care professionals about the lives of children and young people with life-limiting conditions, due to the very small population of this patient group (Craft and Killen, 2007). The structure is therefore geared towards providing the reader with a real flavour, through the words of parents and young people, of the complexity of decisions about technological support. Presenting the model early on also enables the reader to refer back to the model when reading about its different components.

The decision to merge the accounts of parents and young people is firstly related to the structure of the data, with analysis focusing primarily on the decisions within the data. Secondly, one of the emerging themes during analysis regards the shared process of decision-making that happens within the family unit, which was relevant for all the young people taking part in the study, despite having reached the legal age at which they are viewed as independent decision-makers. The interaction

between parents and young people is a key element of the decision process, and viewing their accounts separately risks losing this important feature in decision-making.

The strength of this finding cannot be emphasised enough here, and where parents and young people of the same family took part in the study, their accounts were remarkably similar, even down to the phrases and information they recalled about the decision under discussion. In presenting the findings, the researcher therefore employs the term 'family' to include the unit of the ill child or young person, and his/her parent/s or carer/s. Where the findings presented relate specifically to parents or young adults, these separate terms are applied.

The inclusion of raw data, in the form of direct quotations, was deemed essential in order for the reader to be able to understand how the decisions made by families fit the model. It was also deemed appropriate given the finding in the study that healthcare and other professionals must take the time to understand the 'bigger picture' when they are involved in decisions about technological support and other interventions recommended for children with life-limiting conditions, in other words that they see the child and family rather than simply the condition and treatment. Keeping the research close to the data enables the target audience to do this, as the model is integrated within families' narratives.

The use of pseudonyms and the decision to present the findings in the present tense is underpinned by the same aim, to locate the reader within the lives of families as they make complex medical decisions. There is a danger that by displaying the data in the present tense, it implies to the reader that the findings are being generalised in doing so. However, this is not the intention of the researcher, nor the aim of the study, and a critical reader should be able to avoid making these assumptions. What it does do though, is shift the impact of the findings on the reader, because it suggests that the issues being raised are current and real, and not simply grounded in the past reflections of families about decisions no longer of consequence.

- CHAPTER 5 -

THE DECISION TO INITIATE TECHNOLOGICAL SUPPORT

Following on from the methodology, the next five chapters contain the main findings of the research. Before presenting the new framework of treatment decision-making for children and young people with life-limiting conditions developed in Chapter Six, this chapter introduces the reader to the study sample and data. The chapter begins with an overview of the study sample and a table providing the main characteristics of the child and the decisions made. The remainder of the chapter contains five contrasting case studies to give the reader a flavour of the narratives that make up the data, and to portray the complexity and diversity of the decision processes examined in this research.

5.1 The Study Sample

The sample is comprised of nineteen families in which a child has been diagnosed as having a life-limiting condition. Among the children and young people there are thirteen males and six females with a combination of diagnosed and undiagnosed life-limiting conditions across ACT categories 2, 3, and 4 (ACT, 2012). Although gender appears to be biased towards males, this reflects the wider demographics of children's hospice users (Taylor et al., 2010b). The age group of the children and young people ranges from one year to twenty-three years at the time a decision about technological support was made.

The families vary by structure (see Table 3 on page 126 for information about each family), and the sample contains fourteen two-parent families, one step-family, and four single parent families (one through bereavement, one lone parent, two by divorce). The sample contains one adoptive family. The sample does not contain a family in which there is more than one life-limited child. However, the sample does contain families with varying numbers of children. Of the young adults one lives independently with the assistance of carers, one has lived independently but currently lives at home with his family, and the remaining three continue to live in

the family home. Of the young people living at home, two are cared for by a parent and two have independent carers.

Seventeen families are White British and two families are of South Asian background, reflecting the ethnic make-up in the region from which the sample was drawn (Taylor et al., 2010b). The age range of parents is 32 to 76 years. At the time of being interviewed, twelve parents were in full-time employment, one was self-employed, three worked part-time, and nine were not employed. Educational background, occupation, and level of neighbourhood deprivation varies across the sample, which ranges from the 4th to the 98th percentile on the Index of Multiple Deprivation rank score (using the 2010 English Indices of Deprivation, Department for Communities and Local Government, 2011).

The sample was drawn from a large region in northern England crossing several Local Authority and NHS Trust boundaries, and the sample contains families living in rural areas, villages, towns, and inner cities. All families had accessed support from the recruiting organisation at some point during their child's illness and for some of the bereaved parents also after their child had died. However, seven of the nineteen families did not have access to that provision at the time of making a decision about technological support. Similarly, families in the sample varied in the level and type of statutory support being provided at the time of the decision.

Of the nineteen children and young people in the study, eight had been involved in decisions about gastrostomy insertion and assisted ventilation, eight in decisions about gastrostomy, and three in decisions about ventilation. Among the eight families involved in both decisions, two families opted to focus on only one of these experiences. Therefore in total, twenty-five decisions about technological support are included in the study, and form the focal point for analysis.

Of the twenty-five decisions, fifteen are about gastrostomy and ten are about assisted ventilation. Within the gastrostomy decisions, four families chose not to proceed with a gastrostomy although one young person chose instead to continue being fed by nasal gastric tube. Of the ventilation decisions, only one family chose

not to initiate ventilation should it be needed. Across the sample are examples of decisions made within different institutional settings, different family and medical contexts, and across differing timescales.

Although the data were analysed together to explore the decision-making process, a distinction is made between families of children and young people with no or very limited decision-making capacity, either due to age, maturity or mental capacity, and families of children and young people with developing or full decision-making capacity. Ten families form the first group and nine families make up the second group. The rationale for paying attention to these sub-groups relates to the unique dynamics of decision processes that primarily involve the parent and medical professional dyad, compared to decision processes that involve the young person, parent and medical professional triad.

Table 3 on the pages that follow provides the reader with a summary of each child and family, and the decisions that comprise the data. The socio-economic status, occupation, and ethnic background of individual families are not provided here because of the risk that families could be identified.

Table 3 – Overview of the Sample Decisions

** The names of children and young people are not their own, and have been chosen by the researcher*

Child or Young Person	Decision	DECISION OUTCOME
CHILDREN AND YOUNG PEOPLE WITH NO CAPACITY FOR DECISION-MAKING AT TIME OF DECISION ABOUT GASTROSTOMY OR VENTILATION		
<p><i>Steven *</i> Male age 22 with severe cerebral palsy, epilepsy, and multiple disabilities No communication and severe developmental delay Lives with parents (Steven is adopted). NO TECHNOLOGICAL SUPPORT</p>	<p>Gastrostomy recommended by speech therapist and dietician 3 YEARS AGO</p>	<p>NO</p>

Child or Young Person	Decision	DECISION OUTCOME
<p>Hollie Female age 8 with rare infantile spasm condition No communication and severe developmental delay Lives with parents and older sibling NO TECHNOLOGICAL SUPPORT</p>	<p>Gastrostomy recommended by child's paediatrician 5 YEARS AGO</p>	<p>NO</p>
<p>Josh Male age 8 with diagnosed muscular degenerative disorder No signs of developmental delay * Lives with parents and younger siblings (although in PICU at time of decisions) 24 HOUR VENTILATION BY TRACHEOSTOMY GASTROSTOMY FEEDS OVERNIGHT * Josh was age 1 and 2 when the decisions were made and therefore had no capacity for decision-making at the time</p>	<p>Ventilation recommended by PICU consultant 7 YEARS AGO</p>	<p>YES</p>
	<p>Gastrostomy recommended by PICU consultant 6 YEARS AGO</p>	<p>YES</p>
<p>Emily Female age 15 with diagnosed neurological disorder No communication and severe developmental delay Lives with parents and older sibling GASTROSTOMY FEEDS OVERNIGHT</p>	<p>Gastrostomy recommended by child's paediatrician 7 YEARS AGO</p>	<p>YES</p>
<p>Robert Male age 15 with severe cerebral palsy and epilepsy No communication and severe developmental delay Lives with parents and younger siblings GASTROSTOMY FEEDS DURING DAY (mealtimes)</p>	<p>Gastrostomy initiated by parents with child's paediatrician 7 YEARS AGO</p>	<p>YES</p>
<p>Charlotte Female age 10 with severe cerebral palsy and epilepsy No communication and severe developmental delay Lives with parents and two younger siblings GASTROSTOMY FEEDS DURING DAY (mealtimes)</p>	<p>Gastrostomy recommended by paediatric consultant 8 YEARS AGO</p>	<p>YES</p>

Child or Young Person	Decision	DECISION OUTCOME
<p>Sebastian Male with rare genetic degenerative disorder No communication and severe developmental delay Lived with parents and younger sibling Died at the age of 10, two years after the decisions about ventilation and gastrostomy were made 24 HOUR VENTILATION BY NASAL MASK GASTROSTOMY FEEDS OVERNIGHT</p>	<p>Ventilation recommended by paediatric nurse 4 YEARS AGO</p>	<p>YES</p>
	<p>Gastrostomy initiated by parents with child's paediatrician 4 YEARS AGO</p>	<p>YES</p>
<p>Molly Female with unknown muscular degenerative condition No communication and severe developmental delay Lived with mother, step-father and younger siblings Father had no involvement in her life GASTROSTOMY FEEDS OVERNIGHT Died at the age of 14, at the same time the second decision was made about her gastrostomy</p>	<p>Gastrostomy recommended by child's paediatrician First decision 8 YEARS AGO Second decision 3 YEARS AGO</p>	<p>YES</p>
<p>Lewis Male with severe cerebral palsy and epilepsy No communication and severe developmental delay Lived with parents and older sibling GASTROSTOMY FEEDS OVERNIGHT Died at the age of 3, one year after the decision about a gastrostomy was made</p>	<p>Ventilation discussed with various professionals THROUGHOUT LIFE</p>	<p>NO</p>
	<p>Gastrostomy recommended by PICU 5 YEARS AGO</p>	<p>YES</p>
<p>Casie Female with unknown muscular degenerative condition No communication and severe developmental delay Lived with parents and younger sibling GASTROSTOMY FEEDS OVERNIGHT AND DURING DAY Died at the age of 3, two years after the decision about a gastrostomy was made</p>	<p>Gastrostomy recommended by an unknown hospital paediatrician 4 YEARS AGO</p>	<p>YES</p>

Child or Young Person	Decision	DECISION OUTCOME
YOUNG PEOPLE WITH DEVELOPING OR FULL CAPACITY FOR DECISION-MAKING AT TIME OF DECISION ABOUT GASTROSTOMY OR VENTILATION		
<p><i>Kate</i> Female with diagnosed neuro-degenerative disorder Normal development but loss of communication Lived with mother, two older siblings who lived elsewhere, father deceased NO TECHNOLOGICAL SUPPORT Died at the age of 18, one year after she made a decision about artificial nutrition</p>	<p>Gastrostomy initiated by parent and young person 4 YEARS AGO</p>	<p>NO</p>
<p><i>Peter</i> Male with diagnosed muscular degenerative disorder Normal development Lived with parents and younger sibling OVERNIGHT VENTILATION BY NASAL MASK Died at the age of 20, one year after a decision about assisted ventilation was made</p>	<p>Ventilation recommended by hospice doctor 6 YEARS AGO</p>	<p>YES</p>
<p><i>Lee</i> Male with diagnosed muscular degenerative disorder No developmental delay Lived with mother and older sibling Father no involvement for a number of years NASAL GASTRIC FEEDS DURING DAY (Meal times) OVERNIGHT VENTILATION BY NASAL MASK Died 2 years ago, one year after Lee made a decision about a gastrostomy</p>	<p>Ventilation recommended by respiratory consultant 7 YEARS AGO</p>	<p>YES</p>
	<p>Gastrostomy recommended by main paediatrician 3 YEARS AGO</p>	<p>NO</p>
<p><i>Matthew</i> Male age 23 with diagnosed muscular degenerative disorder No developmental delay Lives with mother, has older sibling who lives locally, father has no involvement in Matthew's life OVERNIGHT VENTILATION BY NASAL MASK</p>	<p>Ventilation recommended by hospice team 3 YEARS AGO</p>	<p>YES</p>

Child or Young Person	Decision	DECISION OUTCOME
<p>Harry Male age 24 with diagnosed muscular degenerative disorder No developmental delay Lives with parents and has older sibling who elsewhere 24 HOUR VENTILATION BY TRACHEOSTOMY</p>	<p>Ventilation recommended by chest specialist doctor 9 YEARS AGO</p>	<p>YES</p>
<p>Sam Male age 16 with diagnosed muscular degenerative disorder Normal development Lives with parents and older sibling GASTROSTOMY FEEDS OVERNIGHT OVERNIGHT VENTILATION BY NASAL MASK</p>	<p>Gastrostomy recommended by main paediatrician 2 YEARS AGO Also involved in decision about ventilation</p>	<p>YES</p>
<p>Alex Male age 22 with diagnosed muscular degenerative disorder No developmental delay Lives with mother and two younger siblings although has lived independently and spent long periods of time in hospital Father has some involvement in his life GASTROSTOMY FEEDS OVERNIGHT 24 HOUR VENTILATION BY TRACHEOSTOMY</p>	<p>Gastrostomy recommended by intensive care doctors 2 YEARS AGO</p>	<p>YES</p>
	<p>Ventilation recommended by intensive care doctors 2 YEARS AGO</p>	<p>YES</p>
<p>Richard Male age 22 with diagnosed muscular degenerative disorder No developmental delay Lives independently with carers Parents and younger sibling live close by OVERNIGHT VENTILATION BY NASAL MASK GASTROSTOMY FEEDS DURING DAY</p>	<p>Gastrostomy recommended by main paediatrician 6 YEARS AGO</p>	<p>YES</p>
	<p>Ventilation recommended by chest specialist doctor 9 YEARS AGO</p>	<p>YES</p>

Child or Young Person	Decision	DECISION OUTCOME
<p><i>Simon</i> Male age 32 with diagnosed muscular degenerative disorder Normal development Lives with parents and has older sibling who lives locally 24 HOUR VENTILATION BY NASAL MASK</p>	<p>Ventilation recommended by chest specialist doctor 9 YEARS AGO Also involved in decision about gastrostomy</p>	<p>YES</p>

5.2 Introducing the Case Studies

Five contrasting case studies are presented here, selected to capture the complexity of decision-making, and to draw attention to the interplay of factors regarding decisions about technological support, the unique family dynamics and circumstances, and the influence of professionals and the wider narrative around a child’s care and treatment. The cases provide an insight, through the words of families, into the complex and individual journeys families take in making decisions for their ill child.

The accounts are descriptive, with reference to the raw data in order that the reader can familiarise themselves with the data before considering how it has been interpreted by the researcher in moving towards a new model of decision-making. The case studies introduce the key themes identified during analysis, drawing out in particular the complexity and fluidity of quality of life; the inherent uncertainty involved in decisions about technological support; the interplay between choice and decision-making; and the essential role of information and opinion in reducing uncertainty and enabling families to make the right choices.

Lee’s story is told by his mum, although her narrative focuses on Lee’s involvement in the decision he made to forgo a gastrostomy. Matthew and his mum provided separate accounts of the decision about assisted ventilation. For Sam and Emily, both parents shared in the narrative about the decisions they made regarding a gastrostomy. Although the severity of Emily’s cognitive impairment means that her

participation in decision-making was not possible, Sam's voice is missing from the narrative, and this is partly due the way in which his parents view his ability to cope with and understand the decisions that affect him. In contrast, Sebastian was not able to be involved in decision-making, and his story is told by his dad, who gave up work to become the primary carer after his son was diagnosed with a progressive and life-limiting condition.

Here follow the stories of Lee, Emily, Sam, Sebastian and Matthew.

5.3 Lee's Decision

Lee, who died unexpectedly at the age of 16 from a muscular degenerative condition, was fed by nasal gastric (NG) tube for eight years before he died. At the age of 14, approximately one year before his death, Lee chose not to have a gastrostomy after it was recommended by his consultant. Both his consultant and his mum, as the two adults who had regularly been involved in choices about his condition and care over the years, believed that Lee would benefit from a gastrostomy.

Nevertheless, Lee's decision was respected, and the reasons for his final decision remain unknown.

"He didn't want it done and that was fine. So that was, and that was it. But nobody ever, after that, said to him, you know, they spoke to me and said "was there a reason?" I said, "I just don't know"."

Lee's mum on the uncertainty she felt about Lee's decision¹

In Lee's case, one of the main factors that his mum believed influenced both the ultimate decision not to have a gastrostomy, and the process of making that decision, related to the fact that Lee had already adapted well to living with an NG tube, despite the negative experience mum described when it was first initiated.

Footnote

¹ The notes placed under each quotation are provided to guide the reader, where needed, to the case from which the passage has been drawn, and offer some of the wider context around the point being illustrated. They are additional to the findings presented and are not required reading. However, they may be useful to the reader in locating the case, and considering how the case illustrates the points being presented.

“He used to hate it, hate it, hate it, and to have it taken out, that was horrible, removed, you know when it blocks, so you had to remove it, and then you’d have to, and then to pass another one, oh it was awful.”

Lee’s mum talked about the early days of having an NG tube.

“Well he got so used to it [the NG tube] he knew it was a means to an end”

Lee’s mum talked about the NG tube as time passed.

When a gastrostomy was first suggested, Lee’s mum reported that Lee had no intention of considering it, and was happy with his NG tube.

“The gastrostomy actually, oh they talked about it. He sort of like, “no it’s alright. I like the [NG] tube thank you very much. It’s alright, I don’t want it”.”

Lee’s attitude towards the gastrostomy when it was first mentioned.

What’s more, mum had made changes over the years to minimise the risk of blockages and other complications associated with the NG tube, and also to reduce its invasiveness aesthetically.

“And then I discovered flesh coloured tape...Ah it looked brilliant. You could hardly see it and I think that’s what helped him because of, so that people didn’t initially say “what’s that tube up your nose for”.”

Mum’s attempts to minimise the invasiveness of the NG tube for Lee.

The fact that Lee had been fed via nasal gastric tube for a long time also meant that the separate decision for Lee to be fed artificially had been made many years before, and the distress mum experienced in accepting the loss of oral feeding for Lee happened at the time of this first decision.

“They said “right we’ve come to a point. We’re going to have to have an NG tube”. Now, I argued the toss for years, sort of, “you said it wouldn’t last this long”. It wasn’t gonna last this long, you know, with an NG tube. But what they said was basically it was going to be for a short time. Now

in my head I thought short time was only going to be for six weeks.”

Lee’s mum found it difficult to accept artificial nutrition would be required long-term.

For Lee and his mum, the decision about a gastrostomy therefore involved weighing up the benefits and drawbacks of two different options – using a gastrostomy, ‘the unknown’, and using an NG tube, ‘the known’. Lee therefore perceived the decision about a gastrostomy as a real choice between two alternatives, and the background to the decision was particularly relevant because it offered an understanding of Lee’s life with an NG tube.

“I decided that I just felt, well if he was still comfortable with the [NG] tube. It was a choice thing, coz I’d felt that he was still fine with the NG tube. It was perhaps a visual, a cosmetic issue that he might need to change.”

Lee’s mum highlighted the choice Lee had to make between two viable options.

Because Lee was comfortable with his NG tube, and was already receiving adequate nutrition, the decision was not time sensitive, and Lee was therefore able to deliberate over the decision for many months. The professionals involved in the decision were also supportive of this approach, and mum highlighted their acceptance of the NG tube as a viable option over the long-term for Lee, despite its general use for patients as a short-term intervention.

“No they didn’t say, “Right this is the end of it. I think what we need to do is give you a gastrostomy”. It wasn’t like an emergency gastrostomy or anything like that.”

Lee’s mum praised the approach by professionals to give Lee time to decide.

Because the decision was not time sensitive, Lee was able to gather more information than would have been possible if a decision had to be made quickly. In addition, Lee and his mum had developed good relationships with a number of professionals over the years, so they were able to obtain more than one opinion, and access information not provided at the point of recommendation.

“He wanted to have a look at every way of what it looked like, how they would do it, who would do it, how they would put, what sort of masks they would have on if he had to go to sleep you know, what was the process.”

Lee’s desire to make a fully informed decision about a gastrostomy.

As well as talking to more than one consultant, Lee spoke to another young person with a gastrostomy to find out what it was really like to have something in his stomach. Lee also made attempts to collect information about how it would affect his own personal circumstances, in other words to individualise the risks and benefits.

“He wanted to know basically whether his trousers were going to rub on it, whether his brace would be messed up, you know would he still be able to sit up as well because his brace would be messed up because it would have a bloody great hole in it.”

Lee’s mum talked about the information Lee wanted in order to make a decision.

The possible complications and drawbacks were important factors, and Lee sought advice from a respiratory consultant because of the risk his respiratory problems posed for surgery. His mum too considered the possible complications, and recalled her encounters over the years with other families whose child had a gastrostomy.

“I’d seen some really gruesome gastrostomy problems, you know, while we were in hospital, do you know what I mean? Not gruesome, that sounds awful, but you know some of the, how sore they were, or how they’d become, because they weren’t perhaps bathed or cleaned properly or whatever.”

Lee’s mum worried about the complications of having a gastrostomy.

The uncertainty regarding Lee’s condition, and the risks it posed for the outcomes of having a gastrostomy, meant that possible complications were considered carefully. In this regard, the uncertainty of a gastrostomy compared to the familiarity of the NG tube was a difficult factor for Lee and his mum to overcome,

particularly as the 'unknown' option was also the option posing a greater risk for Lee, due to the need for surgery and the uncertainty about whether or not the gastrostomy would be inserted at all.

"It was quite hard hitting, it was quite honest questions, that actually you know it might be that, [now referring to the consultant] "I've got to be honest with you Lee. I might actually put you under anaesthetic and actually, we might not be able to do it because, can you see from these x-rays, you know, some of your bits aren't in all the right places"."

Lee's mum highlighted the additional risk of having a gastrostomy for Lee.

Lee's prognosis regarding his condition, and his quality of life at the time a gastrostomy was recommended also influenced his decision not to proceed. The risks and complications associated with the gastrostomy were weighed against Lee's existing quality of life, which was viewed positively by Lee and his mum. 'Wasting time' having surgery and recovering would mean taking time away from 'living' at a stage when life was still busy with school, friends, and other activities and hobbies. Although the gastrostomy had been planned for the school holidays because Lee was keen not to miss any school, he did not want to miss out on the summer holidays either, particularly as there was no 'medical need' to do so because of his NG tube.

In retrospect, Lee only had one more summer after this one, and his pursuit to live a full and active life had been something Lee's mum had strived to achieve since Lee was diagnosed as a toddler.

"You know, god forbid that you lose out on any time in the holidays for goodness sake. And actually, in retrospect, yeah, he was right."

Lee's mum reflected on Lee's decision to enjoy life rather than take time out for surgery.

As well as not wanting to miss out on 'living', mum believed that Lee chose not to proceed with a gastrostomy because of the fear that something might go wrong,

which she explained was caused by the uncertainty regarding his condition and the unknown outcomes of having a gastrostomy.

“We never properly got to the route of it all...whether or not he had a vision and he knew he wouldn’t have come through it, whether he just started to panic and thinking things were going too fast, which I think was more likely, you know what I mean?”

Lee’s mum revealed Lee’s fears about having a gastrostomy.

Finally, the close relationship between Lee and his mum, and her respect for his growing independence and capacity for decision-making, enabled Lee to take responsibility for decisions about his life, and ultimately to make a decision that went against what was being recommended by his consultant and his mum. In acknowledging Lee’s desire and ability to make his own decisions, mum assumed responsibility for ensuring he could make a decision that was fully informed, and arranged an array of meetings and consultations during which Lee was able to obtain information and advice about the decision he would make.

“I think that they [referring to young people] should be involved in every angle, or every aspect of decision-making, but to find out as much as they possibly can if they want to.”

Lee’s mum talked about her role as information gatherer for Lee.

Mum also made herself available to discuss new information with Lee, and answer any questions he might have, and knowing that Lee was fully informed and made his decision on this basis, helped mum to accept his decision and consent on his behalf.

5.4 Emily’s Decision

Emily, who has a diagnosed neurological and developmental disorder, and suffers from severe epilepsy, was 8 years old when a decision was made for her to begin feeding by gastrostomy. Due to her condition, Emily suffers from severe cognitive and communication impairment, and the decision was therefore made by the

parent-professional dyad. Although Emily's potential for participation in decision-making was severely compromised, her life before the gastrostomy, and her quality of life at the time of making a decision, were still central factors in the decision process her parents engaged in at the time.

Unlike Lee's decision though, the main factor identified by Emily's parents, both of whom took part in the study, was the perceived lack of choice regarding their daughter's gastrostomy.

"You haven't got a choice. Well you have, but if we hadn't have done what we've done she wouldn't be here now. For whatever reason, whether it's her chest, coz she were 'fitty', coz of her chest, coz she were undernourished."

Emily's dad talks about the lack of perceived choice in the decision.

In addition, the decision for Emily was not just about a gastrostomy, it was also about having to accept the need for artificial nutrition, and therefore accept the loss of oral feeding and what that would mean for Emily and her family. The loss was significant for mum, whose role in feeding her daughter had previously structured each day, and provided opportunities for interaction and bonding during the lengthy mealtimes they spent together.

"The day that she went into theatre and had it done, when she came out of theatre I just couldn't stop crying because I lost another part of my duties if you like, that personal bit of feeding."

Emily's mum recalls the loss she experienced when her daughter had a gastrostomy.

This was a family, that despite their daughter having feeding difficulties for a number of years, had not realised how severe those problems were, and had not in any way anticipated that she may at some point not be able to feed orally. Although Emily had regular check-ups, the consultant had relied on her parents' accounts of her feeding patterns, and consequently assumed there were no problems.

“Well, she were eating a lot. And every time we went to see her paediatrician, is she eating? Yes, she ate like a pig.”

Emily’s mum recalls informing the paediatrician that Emily had no eating problems.

Although Emily could only eat pureed food, mum did not find this unusual and referred to other children at Emily’s school who were the same. On reflection, mum and dad realised that Emily did indeed have difficulties, and they talked about Emily’s health and wellbeing before having an NG tube, and highlighted how thin she had become and the dark circles around her eyes. However, at the time they were unable to see this change in Emily.

Because her feeding difficulties had gone unnoticed, the family did not have access to a dietician or nurse specialist, or other professional who may have been able to help the family become aware of Emily’s deteriorating health state, and the symptoms she showed of aspiration. Her parents admit that at the time they had limited knowledge of their daughter’s condition, and had not realised that feeding difficulties were often a problem for children like Emily.

Dad: “We didn’t realize. We thought, oh it’s just a bit of food. It’s got in with her snot basically. We were thinking things like that. And it were getting worse wasn’t it?”

Mum: “Yeah but because she were eating it didn’t matter.”

Emily’s parents talk about not realizing how serious Emily’s feeding problems were.

It was only when Emily’s paediatrician saw Emily being fed during a regular check-up that it became apparent Emily was aspirating.

“We went to see the paediatrician and I says, oh she’s due a feed. So I got a couple of yoghurts out, and it came down her nose and she coughed, which she normally did. She went, “don’t feed her no more”. That were it. NG tube down.”

Emily’s mum recalls being told to stop feeding her by the paediatrician.

The rapid circumstances that led to Emily being fed by nasal gastric tube, and the finality of being told that Emily could not be fed orally due to the risk it posed to her health was a shock for the family, and the decision about artificial nutrition was taken out of their hands. However, the NG tube gave mum and dad time to get used to the idea that Emily needed a gastrostomy. The hospital also ran tests during this time to confirm that Emily was aspirating. Furthermore, because the benefits of the NG tube were noticeable after only a few weeks, they were able to see the advantages for Emily of being fed artificially.

Mum: "But within weeks she started to put weight on."

Dad: "She were different as well because she were getting her medication wasn't she?"

Mum: "Yeah, she were getting the full dose."

Emily's parents discuss the immediate benefits of the NG tube.

As well as putting weight on, Emily's fits had reduced because she was receiving the correct dose of medication. In addition, she was less constipated because she was now receiving more fluid than the spoonful mum reported her to take each day prior to being fed artificially. Mum and dad also talked about Emily being happier and calmer too, which mum believed was "because she wasn't hungry. So she didn't have belly ache".

The period of time between the NG tube and a gastrostomy spanned two months, and this time enabled mum and dad to be more involved in the decision about Emily's gastrostomy than they were able to regarding her NG tube.

"I mean it's not as if like, she needs it now, this second. You've got time to think about it."

Emily's dad was grateful for the time they were given to think about the gastrostomy.

This time was important because Emily's parents knew very little about gastrostomies, partly because they had not anticipated or even considered that

Emily would no longer be able to feed orally. Their knowledge of the associated risks and complications of a gastrostomy was therefore limited.

“We didn’t know anything about it. We didn’t really know any kids that had had it, and if we did we didn’t know much, you know what I mean?”

Emily’s dad recalls not knowing what a gastrostomy was.

Although they were now aware of the benefits of artificial nutrition because of Emily’s NG tube, a ‘gastrostomy’ was still an unknown entity, and being able to see another child with a gastrostomy helped them to understand how it worked, and what it would be like for Emily. For mum, it had been difficult to make sense of how the “bits of plastic with clamps and a bit of tube” worked, and seeing another child helped to reassure her that Emily would be fine with a gastrostomy.

“There was a mum, and her baby had just had it done. And while she were in hospital they asked her if I could go and have a look because I’d never seen one. And we went into this room, and we thought “ooh, a bit of a triangle and a bit of a tube. That’s it”.”

Emily’s mum talked about seeing another child with a gastrostomy.

As with Lee, the fear of surgery was something Emily’s parents experienced, and these feelings were very much part of the decision process.

“It were like there were no choice. It were, oh well, I don’t want her to have an operation. You don’t want your kids to have operations, to stop in the hospital.”

Emily’s mum recalls her fear of surgery for Emily.

Although mum described the fear of surgery for her child as ‘normal’, and pointed out that she would experience this with either of her children, the additional risk for Emily of being put under anaesthetic was an important factor in the decision. Weighing this up against the potential benefits of a gastrostomy helped Emily’s parents cope with the uncertainty of something going wrong while Emily was under anaesthetic.

Mum: "You've got to chew it over and think well, she's got to have an anaesthetic for an hour. She's not right good with anaesthetic. She takes a lot of waking up because of [her condition]. So, there's all that but then you've got to think years ahead. She's gonna get all her medicines, fits are gonna be better, she's gonna put weight on, she's not gonna be hungry."

Dad: "It just give her a better quality of life."

Emily's parents talking about the benefits and risks of a gastrostomy for Emily.

Emily's mum also drew upon her views of the wider health care system to help reassure her that Emily 'needed' a gastrostomy.

"I mean they're not going to suggest it if they don't need it. Coz it all boils down to money. They're not going to pay all that money out for, anaesthetist, nurses, and mic-keys."

Emily's mum talks about the influence of her views about the NHS in the decision.

For Emily's parents, the fact that she needed a gastrostomy was never questioned because the risk of aspiration was so severe, and the decisional control assumed by Emily's paediatrician so immediate and clear. However, the process of agreeing for Emily to have a gastrostomy was still important for Emily's parents, because although they felt there was no choice in the decision, it enabled them to weigh up the benefits and drawbacks, and come to their own conclusion that a gastrostomy would improve Emily's quality of life.

5.5 Sam's Decision

Sam, who has a muscular degenerative condition, was, like Lee, 14 at the age of having a gastrostomy, and he too has normally developing capacity for decision-making. However, despite the emerging child-parent-professional triad in this case, the decision itself involved the parent-professional dyad, although Sam was involved in the wider decision process. When asked about Sam's role in the decision, his parents explained that Sam was not able to understand, or cope with

participating in treatment decisions. Consequently, Sam's views were not obtained in the process of deciding whether a gastrostomy was appropriate for him.

Dad: "It was pointless because it would only have worried him."

Mum: "Because he doesn't really understand. I mean obviously we told him he was going for an operation and "it's going to be a tube in your tummy to help put feed in". And he accepted it didn't he?"

Dad: "Yeah, he went along the lines of "I don't want an operation". "No we don't want you to have one either but they recommend it. It's best to have it." "

Sam's parents on the reasons they did not involve him in the decision.

Despite nearing the age at which Sam's consent would be legally required, neither Sam's consent, nor assent, were obtained. Instead, his parents informed Sam that he would be having a gastrostomy, after deciding on his behalf. However, they did at this point provide him with information about why he needed a gastrostomy and what would happen, and offered Sam reassurance that it was "for the best" after he showed initial resistance to the idea and expressed fear about having surgery and being put to sleep.

Again, like Lee, the background to this decision was particularly important because it provides information as to why Sam's parents agreed to a gastrostomy at this time. Although Sam was underweight, it was in fact Sam's upcoming surgery for spinal rods that led to the recommendation for a gastrostomy.

Dad: "He had this operation, his back operation in the March, and in the September before he had his button done. And the reason why he had his button done was to build him up for the [back] operation because he was underweight. They thought if he had his button done and get him fed up..."

Mum: "Then if he's unconscious they can feed him regardless."

Sam's parents talk about the link between Sam's spinal rods and his gastrostomy.

Sam's parents' desire for him to have spinal rods, combined with Sam's paediatrician recommending that a gastrostomy would help to ensure Sam could be fed adequately during this time, led to their decision. However, being informed that Sam could not have spinal rods unless he first had a gastrostomy surprised Sam's parents, who at the time were expecting a date for his spinal rod surgery. In fact, this was the first time a gastrostomy had ever been mentioned by the team involved in Sam's care.

"That came in sudden because we were expecting him to have his back operation. And so then we went through to [spinal operation] hospital and "oh, we're gonna put him in for this instead". And we thought "well what's this?". Then we got told why and the reason what for. "With him being underweight, you'd just find it more beneficial to have it in." "

Sam's dad talks about his surprise when a gastrostomy was first suggested.

Prior to the recommendation, Sam's weight had not been something his parents worried about, and although feeding was becoming more difficult for Sam due to the progression in his condition, he was still able to eat and drink. The family, nor the professionals involved in supporting Sam, were concerned about his nutritional state at the time a gastrostomy was recommended, although Sam's parents believe that it may have been recommended at some point in the future.

Mum: "They would have done eventually."

Dad: "They would have done."

Mum: "They were going to, but they realized that he was underweight, well on borderline wasn't it so they thought the safest thing..."

Dad: "For the operation. Otherwise they wouldn't do it."

Mum: "The safest way was to put the button in so they could still keep feeding him."

Sam's parents discussing the future need for a gastrostomy.

At the time though, the 'medical need' for Sam to be fed artificially was not acute. Nevertheless, Sam's parents felt there was little choice regarding the decision because of the implications for spinal rod surgery if they chose not to go ahead with a gastrostomy. Sam's parents talked about the window of opportunity during which spinal rods could make the most difference for Sam, and the need for his surgery to happen during this time placed them under pressure to agree to Sam's gastrostomy straight away.

Dad: "And you know, we were thinking, "for crying out loud, we'll be having this [the gastrostomy] done now so that's putting it [spinal rods] back". And in between all that, you've got, "it's got to be done when it's right". We got told, for his back. "You let it go too bad..." "

Mum: "Then it's too late."

Dad: "Then you can't correct it. And you're thinking, he was flopping in his wheelchair. He, he would like, he'd be sat up and you'd sit him upright and then he would just go down like that."

Sam's parents highlight the urgency regarding Sam's spinal rods.

Sam's parents recalled a three week period from when a gastrostomy was first suggested to the family, to Sam having the gastrostomy fitted. The decision process itself was very quick, and Sam's parents agreed almost straight away so as not to cause further delays to Sam's spinal surgery. The hospital then contacted them as soon as a slot became available, and in fact the associated paperwork arrived at the house after Sam had been fitted with his gastrostomy, and was in the hospital recovering.

Dad: "Coz I said "if there is a cancellation..." "

Mum: "We'll have it."

Dad: "You know, "Get hold of us and we'll jump in. We can drop and go." Coz we just felt that he was leaving it too late for having his back done

really.”

Sam’s parents highlight the urgency regarding a gastrostomy.

During these weeks, Sam’s paediatrician, along with the surgeon who carried out the gastrostomy and a specialist nurse based at the same hospital, explained all about the gastrostomy and why Sam needed it, and this information was helpful in reassuring Sam’s parents that it would benefit Sam and was worth the extra wait for his spinal rods surgery.

“Because initially you thought “you’re wasting my time”. But when they reasoned and explained it you thought “yeah, good idea”.”

Sam’s dad on the decision process regarding a gastrostomy.

Because Sam was still able to eat, and was deemed ‘underweight’ but not at risk of malnutrition, the decision Sam’s parents made was not about the loss of oral feeding, and Sam’s parents were fully informed that he would be able to continue eating and drinking following a gastrostomy. Instead, the decision was about using a gastrostomy to ensure Sam was healthy and strong for his upcoming surgery. Knowing that a gastrostomy might be needed in the long-term due to Sam’s feeding difficulties and underweight state, also confirmed for Sam’s parents that it was the right thing to do.

Dad: “It was just to boost him up.”

Mum: “Just to give him some extra for his operation.”

Dad: “Boost him up for his operation really, initially.”

Mum: “To get more vitamins in him.”

Sam’s parents talk about why a gastrostomy was needed.

Going through the process of deciding, even when there was little choice, was important for Sam’s parents, who talked about the responsibility they assumed in making decisions for Sam. Knowing that a gastrostomy would benefit Sam was

therefore crucial in helping them consider the risk of complications, and the uncertainty regarding the outcomes of a gastrostomy for Sam.

Dad: "We've had to do it all on his behalf you know. The only thing is if you've got it wrong you're thinking "I hope this doesn't happen". Coz you are told "if it does go, it can this, it can that, it could do that, it might do that. You might this, you might that". And you're thinking "yeah...." "

Mum: "We've made the decision."

Dad: "You know, you're making it on his behalf. And if it did go wrong you know, you've made the decision."

Sam's parents talk about being responsible for decisions about his condition.

Uncertainty regarding the individual progression in Sam's condition was also a factor in the decision process, and being informed about the likely progression was something Sam's dad felt was important when considering the long-term impact of decisions for Sam.

Dad: "They only tell you so much. And I've always said to them, "I want to know what's the next problem gonna be". I don't want to wait till it arrives on my plate. I want to find out before. But they don't always give you everything coz you might not go down that road."

Sam's dad talks about the uncertainty of the future.

Three professionals were involved in information giving for the decision they made – Sam's consultant paediatrician; the surgeon who would carry out the gastrostomy; and a nurse specialist. Each of these professionals provided different types of information, and the nurse specialist was particularly helpful in providing information about what it would be like for Sam, and the impact on his life. In addition, she gave the family written information they could take home, and pictures of how a gastrostomy worked, and what it would look like.

Sam's parents talked about their desire to get information from sources other than the hospital. For Sam's gastrostomy, they accessed information produced by a

condition-specific national charity, and information available online. Sam's parents also drew on their encounters with other children similar to Sam when thinking about the benefits and drawbacks of having a gastrostomy. Their visits to a children's hospice were especially fruitful in this regard.

Dad: "There will become a time where he's gonna be a soup dragon [pureed food]. At a guess. From what I've seen a lot of other boys at children's hospice and that."

Mum: "They're struggling eating."

Dad: "They're struggling eating and that. So I think it will come down to that type of thing."

Sam's parents compare Sam to other boys they have met at a children's hospice.

Sam's parents' desire to obtain a range of information partly stemmed from the distrust they held about the health care system, and the motives of those working within it.

"Well it's nice to see it off somebody, and also see it off somebody that's got nothing to do with it. Coz you never know whether somebody's pulling the wool over your eyes, you know."

Sam's parents highlight their distrust of health care professionals.

This position had not been their default one, and it has been former experiences of making decisions for Sam that has led to Sam's parents questioning the motives of professionals, and realizing that information is sometimes held back from families.

Dad: "And you're thinking, coz I said to them [hospital staff] when A was poorly in hospital, "somebody's just pulling the wool over our eyes aren't they?" "No, I can assure you then aren't." You know, but there was one stage where I thought "I wish they'd keep me in the picture".

Mum: "Are they telling us everything?"

Dad: "I'd sooner know every scenario than have it buried over my head."

Sam's parents discuss their need to be kept informed.

Being fully informed was therefore an essential aspect of the decision process, and the team approach delivered by the three professionals identified by Sam's parents helped them to make an informed decision that the recommendation for a gastrostomy was in Sam's best interests.

5.6 Sebastian's Decision

Sebastian, who died at the age of 10 from a rare genetic condition, was supported by a ventilator and a gastrostomy at the time he died. This case study explores the decision for Sebastian to be supported by a ventilator. The decision to begin ventilation was made two years before his death, and involved his parents and a number of health care professionals with a role in Sebastian's care. Due to Sebastian's severe cognitive and communication impairment, he was unable to participate in decisions that affected him. However, Sebastian's dad described his efforts to keep Sebastian informed of what was happening.

"All the time we talked to him.....so with Sebastian there was a constant stream of carers coming and we had to say to them "look he may not be able to speak, coz he lost that ability. But he knows what is going on"."

Sebastian's dad talks about his commitment to keep Sebastian informed.

Sebastian suffered from asthma, had on-going breathing difficulties, and was over the years, susceptible to chest infections. He had also been hospitalized on several occasions for pneumonia. Like the preceding case studies, the decision background is important because it draws attention to the fact that Sebastian's breathing difficulties gradually worsened over the years, and the decision to begin ventilation was just one decision that Sebastian's parents made with regards his on-going respiratory problems. At the time a ventilator was suggested, Sebastian was on a number of medications and had a nebuliser at home, which had been adapted to

meet his needs. There had also been medical emergencies leading up to the decision that were described by Sebastian's dad as life-threatening.

"So, breathing difficulties gradually developed and there was an occasion where he was upstairs and he was going blue.....she [outreach nurse from children's hospice] dismantled the nebuliser and adapted it, used it on Sebastian.....so we're extremely grateful because Sebastian would have died, probably from heart failure, from not being able to breathe. So we gradually realized there were breathing difficulties."
Sebastian's dad describes Sebastian's worsening breathing difficulties.

Over time, however, Sebastian's parents noticed a change in Sebastian's breathing overnight, and they were unsure of the reasons for this. This development triggered Sebastian's parents to seek medical advice, and this point can be seen as the start of the decision process that led to overnight ventilation.

"It reached the stage where at night he would go very, very blue and his breathing pattern became peculiar. Now subsequently we know that this is 'Cheyne-Stoking', which is a breathing pattern which is associated with people who are dying.....Well we couldn't work out what this was and when the GP came round he said, "well he's just seriously ill"."
Sebastian's dad describes the deterioration in Sebastian's breathing.

Initially, Sebastian's parents were met with reservations about whether anything could be done, expressed by their GP who they believed thought Sebastian was dying. What's more, trying to speak to Sebastian's consultant quickly rather than waiting for an appointment proved very difficult.

"It may well have been that I phoned the consultant, I was told by the secretary of one consultant, "consultants don't talk to the patients. You know, you can't ring up and have an informal conversation". So when you're meeting barriers like that it's difficult. We had a heck of a struggle to rectify, to overcome this problem."
Sebastian's dad highlights the difficulties of accessing medical advice.

Instead, Sebastian's parents turned to the children's community nursing team who had been involved in Sebastian's care for many years, and were much more accessible than Sebastian's consultant. Their experience of working with children with complex health care needs, and their existing relationship with Sebastian and knowledge of his condition enabled them to take action that would lead to a recommendation for ventilation. Taking the lead, the team organised for Sebastian's parents to borrow an oxygen monitor in order for Sebastian to be monitored overnight at home instead of having to be admitted to hospital.

"The paediatric nursing team.....they realized what was happening. And it so happened that not provided by the PCT or the health services, but a grateful parent carer had provided them with a CO² monitor, an oxygen monitor, to measure oxygen. So they loaned it to us to measure his oxygen levels overnight.....Well when we monitored his oxygen levels they were down in the 70's, which is incredibly below, incredibly low. People said "he shouldn't be alive". So that's why he was confused, blue and had this very strange pattern. Well, [now referring to the results of the test] it comes out as a long, like a till, that was sent off to the hospital."

Sebastian's dad describes the action taken by the community nursing team.

The decision process was therefore initiated by the children's community nursing team and Sebastian's parents, who worked together to provide evidence to Sebastian's consultant that something needed to be done urgently. In fact, prior to the test results being sent to the hospital, a decision had already been reached between the nursing team and Sebastian's parents that Sebastian needed overnight ventilation.

Making this decision was easy for Sebastian's parents, who were acutely aware of the seriousness of Sebastian's breathing difficulties and the life-threatening implications of doing nothing at this stage. In fact, given the potential consequences Sebastian's dad described it as a decision with "no choice".

“So in a way, there wasn’t a decision about ventilation. Circumstances decided it for us. But the trouble was, actually getting what was appropriate for Sebastian.”

Sebastian’s dad describes having no choice in the decision.

Given the sense of urgency, the delay in co-ordinating the decision with the consultant was distressing. This process took several months, during which time Sebastian’s breathing difficulties continued. The initial test results provided by the children’s nursing team were lost in the system, and a second overnight test had to be carried out in order for the consultant to agree that ventilation was required.

“Now it may have been August time but it required several people to liaise in order to make a decision about providing oxygen but they were on holiday at different times....and every night Sebastian had this problem so it’s incredibly cruel and reflects this indifferent system of rule following.....Anyway, eventually they all came back, had a consultation. And it was agreed that yes Sebastian certainly did need oxygen.”

Sebastian’s dad talks about the delay in the consultant making a decision.

Once a decision was reached, it then took several weeks to organise the necessary equipment. Again, the children’s community nursing team took the lead in co-ordinating the care Sebastian now required.

“The paediatric nursing team who were brilliant, absolutely brilliant. They were alone in the service providers. If they said something was going to be provided, it happened, 100 per cent. With other people it was delays and arguments, and problems. But they were brilliant....And they put a human face to it, and they often bend the rules in order to make the system work.”

Sebastian’s dad on the support provided by the children’s community nursing team.

The family’s existing and on-going relationship with the team also provided Sebastian’s parents with an opportunity to ask questions about ventilation, how it would benefit Sebastian, and what the potential drawbacks were. However,

because Sebastian's parents felt strongly that ventilation was needed, the complications and risks were not a major factor in the decision process, rather ones that needed to be understood and were then dealt with along the way.

Considering the long-term consequences of ventilation was a factor Sebastian's parents included in the decision. This was underpinned by their knowledge about his prognosis at the time. Sebastian's condition had deteriorated significantly over recent years, and his parents were aware that he was venturing towards the end of his short life.

"With regards to ventilation, the last thing we wanted, and particularly at the end, we did not, I had this sort of nightmare scenario of Sebastian dying in distress and I did not want his last memories of this life if you like to be one of struggling to breathe, and fighting for his life."

Sebastian's dad on the need for ventilation at the end of life.

Like Sam's parents, Sebastian's dad believed that it was important to be fully informed when making decisions on Sebastian's behalf, even when there was little choice. Only then did he feel confident that they were making the right decisions for Sebastian.

"Our focus was, obviously, what was in Sebastian's best interest. And it's sometimes very difficult as an adult to put yourself in the position of a child."

Sebastian's dad reveals how hard it is to make decisions for your child.

To achieve this, Sebastian's dad learned everything he could about Sebastian's condition, and about Sebastian himself. As well as doing research, and reading as much as possible about his symptoms and medications, Sebastian's dad, over the years, developed an acute awareness of Sebastian's wellbeing, and was able to detect when his condition was changing, or when he was unwell.

"We knew something was happening before he actually became ill. So if there was pneumonia there, we knew things were on the move. And we

got things prepared.”

Sebastian’s dad described his awareness about his son’s condition.

5.7 Matthew’s Decision

Matthew was twenty when he was involved in the decision to begin non-invasive overnight ventilation three years ago, and like Sam he has a diagnosed muscular degenerative condition. Matthew made this decision at home following separate discussions with a physiotherapist and doctor at a children’s hospice; Matthew’s respiratory consultant and a nurse specialist based at the hospital overseeing his respiratory problems; and his mum. Matthew views the decision about ventilation as one he shared with his mum, and they discussed the decision openly over a period of weeks, a process which he found particularly useful in helping him to overcome his apprehension about using overnight ventilation.

Matthew’s mum continues to be involved in the management of Matthew’s condition, despite Matthew’s legal status as a competent adult. However, since Matthew was a teenager, his mum has respected his opinions on matters affecting him, and stood by him when at the age of ten Matthew decided against the recommendations being made for him to have spinal rods. Both Matthew and his mum believe that it was Matthew alone who made the final decision about whether or not to initiate overnight ventilation.

Mum: “I’ve always valued Matthew’s opinion and I’ve never gone against what he wants, you know, right from being little I’ve involved him in decision-making and I think you should, you know.”

Matthew’s mum talks about Matthew’s growing role in decision-making.

Reflecting back, Matthew recalled that in the past he had been informed by his consultant that some young people with his condition benefit from overnight ventilation. Staying at a children’s hospice and spending time with other young people who already received overnight ventilation also led him to believe that perhaps one day he might need it. However, the idea that overnight ventilation

might be something for Matthew himself to consider was first suggested by a trained physiotherapist at the children's hospice he visits.

Matthew: "Thinking back, I think it were mentioned to us but not really.....but it wasn't really offered to us. It wasn't like someone said to us, "have you ever thought of going on ventilation to help". Basically it took someone at [children's hospice] to suggest it for us, and for us to ask."

Matthew talks about the beginning of the decision process regarding ventilation.

Following this suggestion, Matthew's mum made an appointment with the hospital to discuss whether Matthew would be considered for assisted ventilation. However, the overnight monitoring showed that Matthew's breathing difficulties were not serious enough to justify support from a ventilator, and Matthew's mum found it very frustrating that the consultant would not consider his other symptoms, and the impact his respiratory problems were having on his everyday life. At this point, Matthew felt unwell every morning on waking, and became very tired in the afternoons, and consequently spent a lot of time at home and in bed during the day.

Mum: "Matthew had needed it for a long time before we actually got it. And we did actually have a bit of a fight to actually get it.....coz they were just using the overnight oxygen levels as an indicator and not looking at the bigger picture of how Matthew was in himself."

Matthew's mum described the difficulties of assessing Matthew's need for ventilation.

The children's hospice was particularly useful during this time because Matthew had only recently made the difficult transition to adult services. Rather than having a paediatric consultant who would oversee his care, Matthew had a number of adult consultants, each with their own specialities. Matthew's mum believed this caused difficulties in obtaining overnight ventilation for Matthew because the consultant involved rarely treated patients with Matthew's condition.

Mum: "Once they've gone to adult services you've got, we go to three different clinics. We see a neurologist in [hospital], a chest doctor in [second hospital], respiratory doctor in [third hospital].....so everybody looks at their own little bit.....It's like you've just been forgotten really, at a time when his needs are increasing. You know, it seems like the actual support, health wise, is less."

Matthew's mum highlights the difficult transition to adult services.

The hospice doctor acted as an advocate for Matthew at this time, and put "pressure" on the consultant to re-consider assisted ventilation. In fact, beginning ventilation at a point before Matthew really needed it was described as an advantage, both for the decision process, and also for the time it took for Matthew to get used to the ventilator.

Matthew: "I felt like it was a choice even when I had it, it was a choice because I didn't need it if that makes sense. It would benefit me but I could survive without it.....because when I first got it I didn't always use it every night.....it was a benefit but it wasn't like, "oh well if you don't use this now you're gonna get really ill"."

Matthew talks about the choice in the decision regarding assisted ventilation.

Mum: "Because there was no emergency for him to be on it. It was better that way, he had more of a choice."

Mum also highlights the choice Matthew had regarding ventilation.

Nevertheless, Matthew did have reservations about using overnight ventilation, which he described as invasive due to the mask he would require.

Matthew: "I remember being a bit apprehensive about it, at first, thinking oh it's, you used to see it and it's just like, you kind of, a mask on my face in the middle of the night. And I thought "it's a bit abrasive and a bit, I don't know, look a bit like some cyborg's attached to you"."

Matthew was apprehensive about using overnight ventilation.

Mum: "I think it were one of those things he didn't want to go on but he knew it were, kind of inevitable, it probably would be eventually. You know, and I mean now he's got quite used to it. I think he felt embarrassed at first with it."

Mum talks about Matthew's embarrassment regarding ventilation.

Matthew also described the anticipated loss of 'normality' he associated with overnight ventilation at the time of making the decision because of the appearance of the ventilator and mask. He described feeling nervous about it, and felt that he would be seen as 'more disabled' with overnight ventilation.

Matthew: "It felt a bit like a step backwards if you know what I mean. As in, coz obviously I'm disabled and I don't really think, try and think too much of, you know, me disability and stuff, and I think it was just one of those things that shows my disability if that makes sense."

Matthew's talked about the appearance of a ventilator and mask.

However, talking these concerns through with his mum and with a friend who also used overnight ventilation gave Matthew a better understanding of how it would benefit him, and helped him to overcome the feelings of apprehension he described. The information provided by the hospital was also useful in helping him weigh up "the pros and cons".

Matthew: "It sort of, well what do you do? I've just got to put these, coz they're silly, behind you, and get on with just getting a better lifestyle."

Matthew describes his mixed feelings about the benefits of overnight ventilation.

The physical feeling of 'needing' more oxygen also helped Matthew to make the decision that using overnight ventilation would benefit him.

Matthew: "I think it was when I was starting to feel really breathless and stuff when I finally decided that I may as well give it a try you know. Sort of like, see how I go on it. It took me a while to get used to it. It took me a good few weeks, maybe months, till I got used to it, and adjustments and stuff. But I've been, like I said, I've been on it 3 or 4 years now and

I'm on it every night now".

Matthew talks about being aware of his need for more oxygen.

The length of time it took for Matthew to get used to overnight ventilation, and the gradual build up, beginning with a few hours each night, or every other night, helped to extend the decision process because Matthew could choose when and when not to use it. However, the gradual approach made it difficult for Matthew to notice the benefits of overnight ventilation at first.

Matthew: "Once I got used to it I did notice the difference. I mean I used to wake up really light headed and tired, and really lethargic and stuff.

And quite quickly, I noticed a difference in general, in mornings."

Matthew highlights the benefits of overnight ventilation.

The decision process also involved Matthew choosing the type of interface he would use with the ventilator – a full face mask or a nasal mask. Matthew was shown both types, and allowed to try them on. "The pros and cons of both different ones" were then explained. However, the main decision to begin overnight ventilation had been made at this point, and this subsequent decision was described as more procedural.

Matthew: "Well that was quite easy. That was basically the same kind of decision as mum saying "what do you want for tea?".....You could try one and if you didn't like that, they'd give you the other one. So it wasn't really like a big issue to choose on that front."

Matthew on the decision to choose a type of mask.

Being able to begin ventilation in the children's hospice was an important factor in the decision Matthew made. His fear of hospitals and his negative experiences of staying in hospital were therefore not factored into the decision because from the outset Matthew was aware he could stay at the hospice. Having access to a children's hospice also provided Matthew with an opportunity to ask other young people about the experience of being on overnight ventilation, and reassured him that over time he would get used to it.

Matthew: "It was like someone who experiences it on the same level rather than someone that might just say try it once, in the training, it's actually someone who actually uses it, uses it on a daily basis. So it was quite interesting to get someone's opinion on it who actually uses it rather than someone that's just, like, they've used it once."

Matthew describes the importance of getting user feedback.

This type of information was important to Matthew, who sometimes found it difficult to understand the technical and medical information provided by his consultant, which he found impersonal because it was about facts and figures, rather than the patient. Like the other case studies, Matthew emphasised the importance of being fully informed, and gathering different types of information. His mum, too, talked about the importance of asking questions, and finding out everything you can, and again like the other case studies, talked about her waning trust in medical professionals.

5.8 Chapter Summary

As the narratives presented here illustrate, no decision is the same, and the circumstances under which decisions are made can be very different. Moreover, the unique combination of child, medical and family factors show just how complex decisions about technological support can be. As well as variation in the information and time families can utilise to make a decision, the backgrounds leading up to the decision can also be very different, some of which can span many years before a recommendation is made for technological support.

The evolving role of families in decision-making for children with life-limiting conditions also has the potential to affect the decision process, as does the interaction families have with professionals and the wider health care system around which decisions are made. Combined, these factors have the potential to influence the way in which decisions are made by families, and can have an impact on the subsequent feelings families might be left with regarding the decision process, whether or not the decision leads to positive outcomes for their child.

The next chapter introduces the new model of treatment decision-making for children and young people with life-limiting conditions developed from the analytical process outlined in Chapter Four. This model captures the complexity of decision-making for life-limited children and young people, and provides a framework from which both the decision and the decision process can be understood.

- CHAPTER 6 -

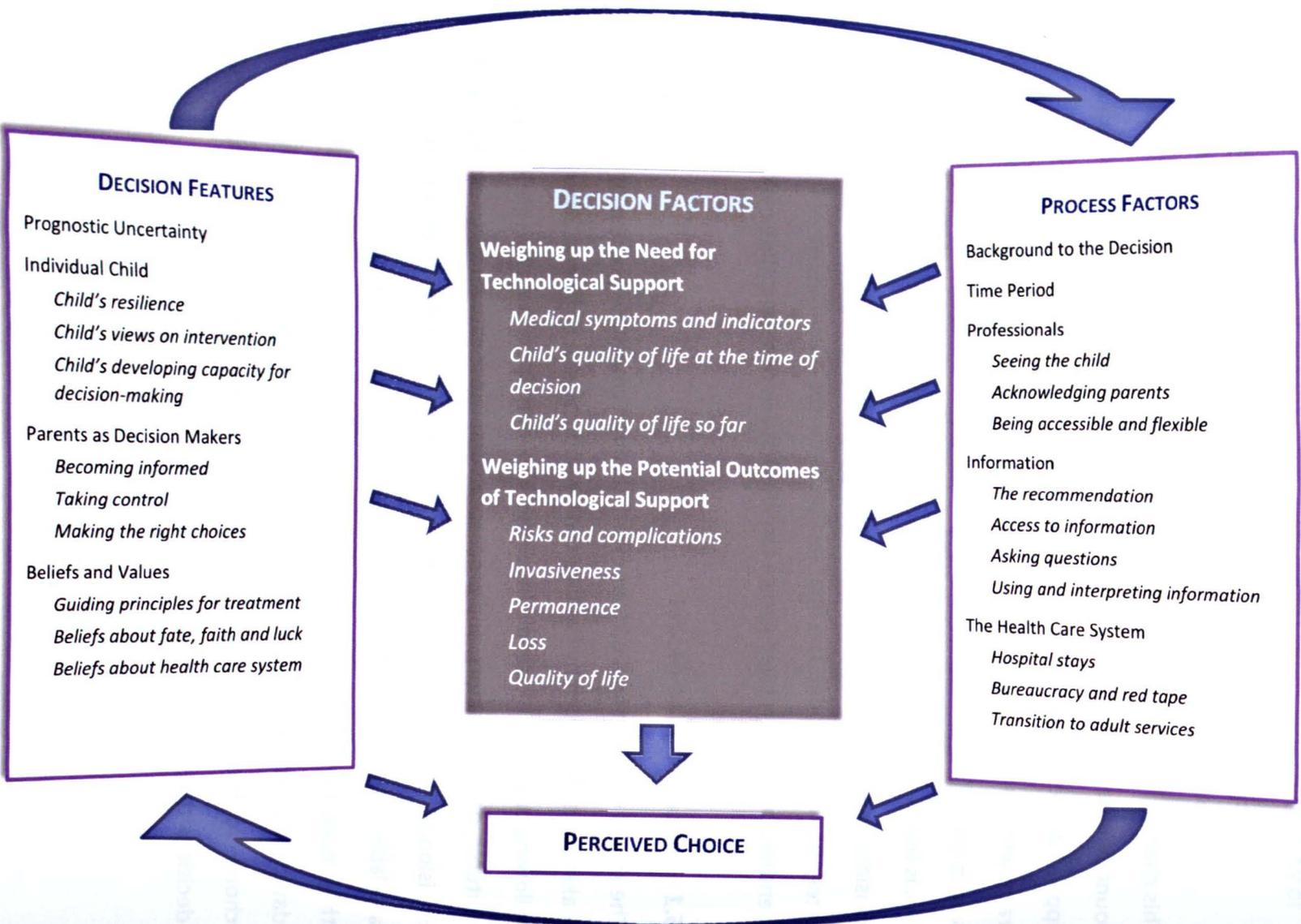
AN ECOLOGICAL MODEL OF TREATMENT DECISION-MAKING FOR CHILDREN WITH LIFE-LIMITING CONDITIONS

This chapter introduces the model of treatment decision-making for children and young people with life-limiting conditions developed from the analytical approach applied to the data presented in Chapter Five. The model is presented as a framework through which to draw together the range of factors that influence the way in which families experience and make decisions about technological support. This is followed with further details about each category in the framework before considering the ecological nature of the relationship between the different categories in the model. The final section discusses the influence of the factors identified on the perceived choice families have in decisions they make.

6.1 Introducing the Model

The model offers an applied framework through which the process of decision-making as experienced by families can be understood. Shown as Figure 1 on the following page, the model distinguishes between three main categories – decision features, decision factors, and process factors. These categories interact during the decision process and can both constrain and enable families as they consider the available options, and affect the perceived choice families have in the decisions they make. Perceived choice rather than the decision itself is a central theme in the data, and relates to the overwhelming desire among families to make the right choices for their child, even when there is sometimes no perceived choice in the decisions they are asked to make.

Figure 1 – An Ecological Model of Treatment Decision-Making for Children and Young People with Life-Limiting Conditions



6.2 Decision Factors

Decision factors, which are those specific to the decision being made, are positioned in the centre of the model and include those factors that parents and young people identify as important when weighing up the appropriateness of the proposed intervention. Quality of life is central to the decision families make, and is assessed in different ways by families as they consider the use of technological support, drawing on their knowledge about their child's life so far, their understanding of their child's quality of life at the time of the decision, and making an assessment of their child's potential quality of life following the initiation of technological support.

Within this category, there is a division between the factors that are used by families in assessing whether or not the difficulties or symptoms their child is experiencing requires medical intervention, and those used in weighing up the potential outcomes of the intervention being recommended. This distinction is important because unlike the latter, which are specifically related to the technological support being proposed, the needs of the child are considered more holistically, and sometimes without the knowledge of the solutions available to meet the needs of their child.

The order in which these sub-categories are presented implies there is a chronology in considering the appropriateness of a proposed technology. In fact, for many families this is indeed the case with parents, as primary carers of their child, being acutely aware that there is a problem, whether or not it has been acknowledged by the professionals involved in their care. However, other families must consider the needs of their child alongside the weighing up of the potential benefits and drawbacks to the technology being recommended. This can occur when the symptoms are not recognised by parents before technological support is recommended, or when there is an acute episode leading to hospitalization and a subsequent recommendation.

Regardless of the circumstances under which a recommendation comes, families emphasise how essential it is that they form their own opinion about the appropriateness of technological support because it enables them to make an informed decision, even when there is little perceived choice in the decision they are asked to make. However, to what extent families are able to engage in the process of considering the needs of their child, and to weigh up the potential benefits and drawbacks to technological support, are influenced by the wider context of decision-making. These are shown in the model as decision features and process factors, which can either enable or constrain families in the decisions they make, and are positioned on either side of the decision itself, with arrows positioned to indicate the influence they can have on the process of weighing up the appropriateness of the technology under consideration.

6.3 Decision Features

Decision features include particular characteristics that are specific to the patient population, in this case children and young people with life-limiting conditions and their families. They are included within a distinct category because they are not about the use of technological support per se, but instead will feature in many of the decisions families make regarding the management of a child or young person's condition. Nevertheless, they influence both the process of decision-making experienced by a family, and affect the way in which decision factors are viewed and understood in the weighing up of a proposed intervention.

Some of the features relate to the life-limiting nature of a child's condition and the prognostic uncertainty inherent in many of the decisions families must make. Others concern the individual child, highlighting the importance of including the child's views about a proposed intervention in the decision process, and considering their ability to cope with the changes that technology will bring to their lives. Gut feelings were also identified as decision features, and the strength and emotional basis of these feelings can make it difficult for families to weigh up the full range of benefits and drawbacks associated with a proposed intervention. These features

draw attention to the emotional context within which these decisions are made, and the overwhelming responsibility parents assume as decision-makers for their child.

Indeed, the role of parents as decision-makers for their child dominated parents' accounts, bringing to light the journey they make from the overwhelmed and naïve parent of a newly diagnosed child, completely dependent on the expertise and knowledge of the professionals involved, to the informed and expert parents they become over time, and the decisional control they assume as they do. The stage parents find themselves at when decisions about technological support are made affects the extent to which they feel able to assess their child's needs, and to weigh up the potential benefits of a proposed intervention. It also reflects the evolving dynamics between parents and the professionals involved in their child's care, and their ability to access the information they require to make an informed decision.

Parents' views on involving their child in decision-making also belong here, as do the experience and views young people themselves hold about the extent to which they should and would like to engage in the process of decision-making. Careful consideration was given to the inclusion of child and parent features within a single category, and the discussion presented in Section 4.7 is relevant here. More importantly though, the family approach underpinning this model reflects the process of decision-making parents and young people engage in, even after they make the transition from child to adult services. Therefore separating out the experience of parents and young people, even when young people have reached an age at which their consent is all that is needed, is not necessarily helpful.

This is a feature that is perhaps unique to children and young people with life-limiting conditions because of their growing dependence on their parents as both their primary carer, their decision-maker, and the individuals in their life who often have the most expertise and individualised knowledge regarding their condition, and how to manage it. The similarity in parents' and young people's accounts of the decision process is striking, and this is also reflected in the beliefs and values they

hold, and through the way they discuss the interpretation of information and opinion for decision-making, and their beliefs and attitudes about the wider healthcare system.

In fact, the views and beliefs parents and young people have about the healthcare system feature in the decisions they make, and are in part influenced by societal and media portrayals of the National Health Service. It was therefore essential to distinguish these beliefs from the more tangible factors associated with making decisions in a health care system, because they are not entirely constructed from those experiences. Finally, other beliefs and values were identified as decision features from the data and while families are not always explicitly aware of their role in decision-making, they nevertheless have the potential to influence the decisions families make for their child. This research draws attention to the way in which views about life and death might shape a family's approach to treatment, and how beliefs about the role of faith and fate can influence how families view the decision being made.

6.4 Process Factors

Unlike decision features, which are about the patient population, process factors refer to the wider context within which decisions are made, in other words the factors around the decision that can either constrain families as they weigh up the proposed intervention, or enable them to make an informed decision. The impact of these factors is important because families can be left with negative feelings when it is felt that the process factors shown in the model have constrained a family's ability to make a good decision.

Access to information and medical opinion provides a good example of the relationship between process and decision factors, and where families are left with unanswered questions, or experience a negative outcome that they recall not being informed about, feelings of regret can surface, followed by anger towards the professionals involved in providing information for the decision. Similarly, where

families are able to access a range of information, and engage in a decision process that involves trusted professionals who may already know their child, or who take the time to understand more about their child's quality of life, the decision process can be one that is reflected on positively by parents, even in those cases where the outcomes are poor.

Included in the category of decision processes are the background to decisions, which vary significantly and give an indication of how the problems leading to a recommendation have developed, and who else has been involved up to this point. For some families, feeding or breathing difficulties can go unacknowledged for some time, causing distress to the child and leading to frustration for parents, whose experience in caring for their child, and the growing expertise they assume as primary carer, is felt to be ignored. This can be seen in contrast to the child whose difficulties have been regularly monitored by their paediatrician or a specialist consultant. This background is therefore crucial towards understanding how a decision may already be 'framed' by a family at the point a recommendation or suggestion is made.

Finally, this category includes factors regarding the involvement of professionals in decision-making and the influence of the wider health care system over the decisions families make. In fact, this aspect of decision-making dominated the accounts of parents, who expressed a range of emotions as they recalled certain events and situations to illustrate the points being made. As well as affecting the way in which families are enabled to participate in decisions for their child, and in particular the information resources and professional expertise available to them, the factors regarding the health care system will already have impacted upon the background leading to a decision, and will continue to influence the decision outcomes and consequences.

6.5 The Interaction between Decision Features and Process Factors

The cyclical interaction of process factors and decision features, which continue both during and outside of the decision process as families interact with professionals and services, portrays the ecological nature of treatment decision-making, and reflects the on-going journey families embark on when a child has been diagnosed with a life-limiting condition and the evolving role of parents and children in decision-making. This iterative process also indicates the changing dynamics in the relationships families establish, in some cases over many years, with the professionals who help to care for their child. Finally, it illustrates how the encounters families have with professionals and services can shape their beliefs about the health care system, which in turn can influence future interactions with professionals who are part of that system.

Careful consideration was given to the layout of the model to ensure that the ecological nature of treatment decision-making was reflected appropriately. Two approaches were considered during the analytical process, initially drawing on the work of Bronfenbrenner (1992), who applies an ecological systems theory approach to the lives of children, and then on the work of Hudson and Lowe (2009), who consider a range of theories and models through which to understand the policy-making process. In particular, this work was helpful in considering whether a top-down approach was more fitting, which implies that families are constrained within the health care system that decisions are made. However, after applying the data to each approach, they were dismissed on the basis that the underpinning assumptions about the interaction between families and the health care system did not adequately match the data under analysis.

Further analytical work showed that in fact while there is an interactional nature to the relationship, they are in essence representing two disparate systems, the family system (decision features), and the healthcare system (process factors), which come together at certain points during a child's life in order to solve a health-related problem, in other words to consider the medical symptoms and indicators a child is

experiencing, and to work together in finding a way to minimise the impact of those symptoms on a child's quality of life. Positioning the categories at either side of the decision therefore portrays this distinct ecology in which treatment decision-making occurs. The directional arrows linking the two categories have been included to draw attention to the on-going influence of each system on the other as families and professionals engage in a series of on-going decisions regarding the diagnosis, management and care of children and young people with life-limiting conditions.

6.6 Perceived Choice

The model contains a fourth element, that of perceived choice, which is included as a separate component in the model in order to reflect the influence of the whole process on the level of choice families believe they have in decision-making. Making the right decision is of key importance to parents, who are acutely aware of the decisional responsibility they carry, referring in many cases to ensuring that the choices they make are in their child's best interests. Young people also highlight the importance of having a choice in the decisions they make, and of making sure they understand why an intervention is being recommended and the impact it will have on their life. Even when parents and young people perceive there to be no choice in the decision they face, there is still a need to understand how the proposed intervention will benefit them if they are to believe they are doing the right thing.

For some families, perceived choice is about the decision itself, and within families narratives there is a distinction between a process of decision-making that involves the weighing up of two viable options, and one that involves forming an opinion that the recommendation for technological support is indeed in the best interests of their child. In the first scenario, the benefits and drawbacks of initiating technological support are compared to the benefits and drawbacks of the status quo, and families will sometimes seek alternative solutions to manage the difficulties associated with a recommendation for technological support. In the second scenario, families describe a decision of little or no choice, because the option of doing nothing is not believed to be a real option. In essence, families

describe a process of making a decision, even though they are sometimes acutely aware during the process that the decision outcome is already determined.

The perceived choice families believe they have is directly linked to the severity of their child's symptoms and indicators, with more severe and obvious symptoms providing evidence that their child needs technological support. While it may be easier for families in this instance to understand why their child needs a gastrostomy or ventilation, it can cause them to believe that the decision they have to make is one with no or limited choice. This scenario also implies that there are no benefits to the status quo, and in fact for families to view the decision as a choice, they must be able to identify at least one benefit to the option of doing nothing, which some parents admit only being able to do upon reflecting back on a decision in which the outcomes have already been realised.

Perceived choice is influenced by the wider context of decision-making too, and the interaction of decision features and process factors. Again information and medical opinion is crucial here, as is the approach taken by professionals as to whether they present a recommendation for technological support as a choice between two options, or a decision that parents must agree to for their child. The level of experience and expertise parents and young people have at the time of making a decision can affect their perception of the choice implied in a recommendation, and their ability to view a decision as one of two viable options. In addition, perceived choice can be influenced by the level of certainty about a child's prognosis, and the shifting expectations regarding quality of life. The beliefs and values held by parents and young people that guide their approach to treatment can also influence how families view the decision.

6.7 Chapter Summary

The model presented in this chapter centres in particular on the ecology of treatment decision-making and offers a new understanding of how families of life-limited children and young people experience the process of making treatment

decisions. The model illustrates how both the on-going interactions families have with the professionals involved in their child's life, and the healthcare system as a whole, can transform decision-making dynamics and either enable or constrain families to make informed choices about the management of their child's condition.

The relationship between perceived choice and the process of decision-making is interwoven within the narratives of parents and young people as they reflect back on the decisions they have made, and is an integral and permeating feature of the model presented here. This is discussed further in the remaining findings chapters, which provide a more in-depth account of the factors used by families in weighing up the appropriateness of technological support, and the features and other factors that can both enable and constrain families during this process.

- CHAPTER 7 -

EXPLORING THE DECISION FACTORS

This chapter provides a detailed account of the category of decision factors shown in the model presented in Chapter Six, in other words the factors identified from the research that relate to the decision in hand, and are used by families as they weigh up the appropriateness of technological support. The chapter contains two main sections, beginning with a discussion of how families weigh up whether or not a child or young person requires medical intervention, and finishing with a discussion of how families weigh up the potential outcomes of technological support should they proceed.

7.1 Weighing up the Need for Technological Support

'Does my child *need* this?' is the question parents strive to answer when they consider the appropriateness of technological support for their child. In fact, decisions are underpinned by their child's needs, and are often described by parents in this way.

"You know she was like a skeleton. She needed it."

Molly's mum emphasises the need for a gastrostomy.

"She had the pneumonia initially, then she had the NG tube in, and then it was clear that she was gonna need a gastrostomy."

Casie's mum uses the term 'need' to describe the decision.

Some parents assess their child's needs before a recommendation is made for a gastrostomy or ventilation, whereas others are not aware that their child may need technological support before a recommendation or suggestion is put to them. In these cases, parents must weigh up the potential outcomes of a gastrostomy or ventilation at the same time as accepting that something needs to be done at all.

Where parents are aware of the need to do something before a recommendation is made, some admit lacking the knowledge and experience about what is required.

“I can remember him sitting on the floor there, and I came in, and he was just like a pile of rags. He was completely collapsed. He wasn’t sitting up. He had so little energy that he couldn’t even sit up properly. And I was very distressed and I said to [Sebastian’s mum], “we’ve got to do something about this”.”

Sebastian’s dad describes knowing that something needed to be done.

For others though, the on-going nature of the problems a child experiences leading up to the decision, and the encounters parents have with other children who have been down a similar path, enables parents to obtain information about the solutions available. In some of these cases parents initiate the decision process and ask for their child to be considered for a gastrostomy or ventilation. At this point they may know little about the risks and complications involved and are driven by the need for something to be done.

7.1.1 Medical symptoms and indicators

Although different types of evidence are drawn upon to assess the needs of their child, parents frequently refer to the physical and medical symptoms that indicate their child’s need for a gastrostomy or ventilation, and these become important factors in the decision. In the case for a gastrostomy, the need is physically evident due to a child’s underweight state, and the symptoms of malnutrition or aspiration they display. For some children who are considered for a gastrostomy, the suffering a child endures during the time leading up to a decision provides clear but distressing evidence that their child needs medical intervention.

“If she tried to drink she coughed to the point of vomiting everywhere. And it just sounded like her lungs were just like a swimming pool. It was horrendous.”

Casie’s mum describes the aspiration her daughter experienced.

For ventilation decisions, increased tiredness, difficulties breathing, escalating chest infections, and episodes of pneumonia, provide families with evidence that assisted ventilation might be beneficial.

“But it was the chest infections that were, you know, the main thing that debilitated him quite badly you know.....so that’s basically why he went onto the assisted breathing.....you see he was struggling, coz they struggle to breath, they’re absolutely knackered, you know they’re tired.”

Peter’s mum on the symptoms he displayed.

In some circumstances, a child’s life is threatened by their feeding or breathing difficulties. An acute episode of pneumonia or an incident during which a child stops breathing due to aspiration or respiratory weakness, again provides a clear message to parents that their child needs something more than they are currently receiving.

“That’s how we realised that Alex could not cope without full ventilation.....we took the mask off and she’d just got it into his mouth and his SATS just dropped and plummeted.”

Alex’s mum describes the incident during which they realised Alex needed a tracheostomy.

For other families the ‘need’ is less pronounced, and the symptoms harder to detect. Although a lack of absolute need can enhance the perceived choice in the decision families make, it can also make it more difficult for families to understand why their child needs support. An example of this relates to young people who begin ventilation at a point before they physically need it, which can make weighing up the benefits against the drawbacks more of a challenge. For decisions about artificial nutrition, some children appear to be coping with their feeding problems, and a recommendation for a gastrostomy can be difficult for parents to accept, who struggle to understand why their child needs artificial nutrition.

“She had a muscle wasting disorder as well so she were really thin, she never put weight on, I mean but she were eating. She could eat bowls

and bowls of liquidised food. She could eat loads.”

Molly’s mum found it difficult to understand why Molly needed artificial nutrition.

In other cases, parents spend a significant portion of each day ensuring their child receives adequate nutrition, for example trying many different textures and tastes, or providing fluid by syringe or teaspoon. While this provides parents with evidence that their child is having difficulties, some parents struggle to acknowledge this because they believe that the role of providing sufficient nutrition belongs to them, and are committed to feeding their child despite the efforts involved. It can therefore again be difficult for parents to accept that their child might need the help of a gastrostomy.

In fact, the concept of ‘need’ is subjective and there are cases in the sample where this subjectivity has led to a difference of opinion about whether a child needs technological support or not. This is in part due to the different ways in which ‘need’ is interpreted, and many parents, although agreeing that health-related symptoms and indicators are key factors in the decision they make, believe the focus within the medical profession on clinical evidence can mean that other factors about a child’s quality of life can be overlooked.

Dad: “And straight away she looks at him..... “This child will aspirate [referring to speech therapist]. He needs a gastrostomy”.”

Mum: “She saw us feeding him like this and she said “it’s not acceptable”..... There’s no problem, well I say no problem. You just need to know how to feed him that’s all. Just now and again he will just cough but you just put a towel over, and he clears it.”

Steven’s parents disagreed with the speech therapist who believed Steven needed a gastrostomy.

In other cases, the need is not directly related to the technology being considered, or relates to a wider range of symptoms caused by a child’s condition. In Sam’s case, the desire for him to have spinal rods at a time when it would make the most benefit created the need for Sam to have a gastrostomy. For Harry, the decision to

have a tracheostomy, although considered with regards to his on-going chest infections, was also about the impact of his respiratory problems on his nutritional health state and Harry's refusal to consider a gastrostomy as an alternative solution.

"Before I had it done I'd lost like three and a half stone in three weeks which is just like a massive drop in weight which is dangerous. And then, they told us also, that with the tracheostomy, it usually makes your appetite come back."

Harry reflects on the need to do something about his dramatic weight loss.

For some children, an overall deterioration in a child's health can indicate there is something wrong, or that it might be time to seek medical expertise. For parents of children who have a well understood illness, they will sometimes know in advance that their child might need a gastrostomy or ventilation in the future, and this knowledge can prompt them to seek advice when there is a marked deterioration in their child's condition. This foresight some families report having due to the knowledge about their child's condition is in stark contrast to children who have no diagnosis, or a rare condition about which little is known, or children who have a condition where the course of progression is variable and difficult to predict.

7.1.2 *Child's quality of life at the time of decision*

Whilst important, the medical symptoms that indicate a child's need for medical intervention are very much viewed by parents within an overall picture of their child's quality of life at the point at which a recommendation for technological support is made. For children and young people whose medical symptoms are not life-threatening, quality of life is sometimes the key factor in considering the appropriateness of a gastrostomy or ventilation, and parents describe the need to underpin decisions by their child's current quality of life at the time of making a decision if they are to consider the potential impact a gastrostomy or ventilation might have.

The intimate and continued relationship parents have with their child enables them to develop an understanding of their child's personality and abilities as well as their condition and associated symptoms. Facial expressions, verbal utterances, physical movements, and even skin pallor and body temperature, become indicators of their child's quality of life, especially for children with severe cognitive or communication impairments, when a child's smile, a hand movement, or the way their face lights up when they visit certain places, are individual indicators that parents draw upon to assess their child's wellbeing.

"I keep saying to her, "we're going to [a place where Emily likes]". And you can just tell. Her eyes might just, but there might be no expression on her face but her eyes will move as if to say "are we?" And as you're getting closer and closer, "we're going soon Emily". And you can tell she's thinking about what you're saying. And she'll start smiling."

Emily's dad describes Emily's response to visiting her favourite place.

In assessing their child's quality of life, parents describe three overlapping yet at times conflicting elements;

- the distress and pain their child endures as a result of their condition and associated symptoms;

"She was clearly struggling in those days, and to see her when she was re-gurgitating, for instance, burning so that all the colour went out of her clothes [from the acid reflux]...just get out of this misery. She was very unhappy, very uncomfortable."

Hollie's mum describes the distress Hollie experienced as a result of acid reflux.

"His mouth was full of sores. He was obviously on antibiotics for it but his mouth was just covered."

Alex's mum talks about the complications of the ventilation mask before he was recommended for a tracheostomy.

"She didn't want to eat anymore. Once she had real difficulty eating she didn't want to. In the beginning I made things soft and tried to get it down but there was so little going down. She drank in the beginning, she could only still do it through a straw but then it would come out of her nose. It wouldn't go down. So, and really she was not, she didn't want that."

Kate's mum describes the distress Kate experienced around eating and drinking towards the end of life.

- their child's potential and desire to engage socially and emotionally with their world;

"She understood everything, even though she couldn't reply, coz she used to make noises and stuff.....I mean she used to stick her foot out to trip her brother up when he were running past her, if she were laid on a bean bag.....it used to make her laugh."

Molly's mum describes the subtle ways in which Molly interacted with her world.

"There's no smiles, there's no fun, there's no giggles, you know there's nothing. She gives nothing out in that sense.....Yet the love that she somehow, you know people who meet her are drawn to her."

Hollie's dad talks about how she interacts with her world, despite the severity of her impairments.

"He can answer with his hand, so sometimes there's more there than what you think. Do you know what I mean? He wouldn't know how to do that would he? He has a sense of humour.....unless he's poorly and then he's crampy. But who isn't when they're poorly?"

Steven's dad describes how Steven engages with those around him.

"Josh's hands tire a lot, he gets very tired quickly. And he loses concentration very quickly as well. Whereas music and football,

that's what he really enjoys. He plays the drums and stuff so that's what his passions are sort of thing."

Josh's mum describes Josh's passions despite the deterioration in his condition.

- their child's 'happiness'.

"She had a good quality of life. They said that she didn't but she did [refers to hospital team that treated Molly at the end of life]. She were always happy. She were always out. She had a better social life than me."

Molly's mum reflects on her daughter's happiness.

"As long as she's content then that is the best that we can actually provide, and she's always content. So, it's only when she has a seizure that she goes into distress but by then she's already switched off.....and so contentment is what we're after."

Hollie's mum describes the importance of making sure her daughter is content.

"He can't walk, he can't talk or do anything. But he's lovely. He's a joy to have, he just smiles. When I get up in the morning and I wake him up, and he looks up and I think good god, he's nothing going for him and he's happy you know."

Steven's dad reflects on Steven's happiness, despite his son's limitations.

"He wakes up every morning happy as Larry. Smile on his face.....he's a happy little soul really."

Sam's dad is reassured to know that despite his limitations, Sam is happy.

Although viewed through the individual lens of a child's condition and impairment, and therefore interpreted in different ways for each child, these three factors form the basis of 'quality of life', and are woven into parents' accounts of their children's lives. Even in cases where the medical symptoms cause significant distress to a child, the other elements of quality of life are still a central concern for parents. Where these elements conflict, it can be difficult for parents to make a decision that

involves improving one aspect of their child's quality of life, while at the same time having an adverse effect on another.

Drawing again on the case of Charlotte, this conflict was central to the decision her parents made because the distress Charlotte suffered as a result of her on-going reflux, and the emotional and social bonding her parents associated to feeding and mealtimes, were both important decision factors. Emily's mum also framed the decision in this way, particularly when for Emily, feeding was perceived as one of the few pleasures she experienced.

"I wanted her to try and have everything. I know, it were just, coz I know she enjoyed her food you know.....she loved, oh, Sunday dinners, and beef stew. She loved everything like that."

Emily's mum talked about Emily's love of food.

Robert's mum described the importance of mealtimes in their family, with the evening meal a symbolic event in their daily lives, the point at which the family still comes together and reflects on the day. That Robert was still able to participate in this important activity was a central concern to his parents at the time it was felt a gastrostomy would be beneficial, because it was a part of the day that enabled him to interact with his family in a way that his parents felt was meaningful and in a way that enhanced his quality of life.

For other families, the decision for a child to have a gastrostomy is made when they are already being fed by nasal gastric (NG) tube. Again, a child's quality of life at this time is a key decision factor, and this very much takes into account the impact of an NG tube, with many parents reporting the distress and discomfort their child suffered due to this method of artificial nutrition. "Getting rid of the NG tube" can in these cases drive the decision to agree to a gastrostomy, which for some families is seen as the less invasive technology.

"When he used to have that tube in his nose and he was always pulling it out, and it was always getting sore and we were always having to put it

back in his stomach. It was horrible. It was just horrible you know, having to go through that every time it came out or dislodged itself. So you know, in that sense, it made having a decision, a PEG or a button, it made it easier really in that sense. Because you didn't want to see that all the time."

Josh's mum talked about the distress caused by the NG tube.

Mum: "Was it 3 weeks with the NG tube, she wasn't getting on with it at all was she? She was vomiting, retching, it was clearly irritating."

Dad: "Yeah, it came out quite a lot didn't it?"

Mum: "Yeah she was vomiting all the time but every time she vomited of course she was aspirating it, and not coughing it out. So in the end we just agreed that the gastrostomy was the best thing to do."

Casie's parents describe the problems with an NG tube.

"He was flailing around all the time, very distressed. He just kept yanking his tube out all the time, so it was a very easy decision because every time he pulled it out, coz it was jejunal he had to have an X-ray to put it back in again. And this great big palaver to get it back in. So to us, it was layman's easy decision to make."

Lewis's parents preferred the option of a gastrostomy to an NG tube.

Alex also reflects on the decisions about both a gastrostomy and a tracheostomy in this way, and describes the experience of being ventilated through a mask as "the worst two weeks of my life".

"I was on a mask ventilator then which wasn't doing much for me.....For two weeks I didn't come off once.....It was like two weeks in limbo land. Well like, it was horrible coz, well I kept having hallucinations."

The experience of non-invasive ventilation drove Alex to accept a tracheostomy.

"I never ate enough anyway, or drank enough. And I had the nasal gastric tube in so, and I didn't like that so I wanted that out. So at the

time that was the main reason, was to get that out."

Alex agreed to a gastrostomy so he would no longer be fed by NG tube.

In contrast, Steven's parents felt that the experience of an NG tube led them to question whether he needed artificial nutrition at all, and provided them with an insight into the potential complications of tube feeding, regardless of the method.

"She says "he wants a gastrostomy". I say "well just let's go through with it". She says "no, he's got to start with, he's gonna have a tube in his nose and the machine". So we had that for three months. Well it were a bloody nightmare.....You should have seen that poor kid with the tube up his nose. He were distraught. He weren't happy, it were terrible."

Steven's mum describes how distressing an NG tube was.

Lee, on the other hand, felt that his existing NG tube had a minimal impact on his overall quality of life, and that the time he would waste in changing to a gastrostomy would mean him losing enjoyable aspects of life during the time it would take to recover and get used to a more invasive intervention. In fact, unlike parents, young people did not use terms such as happiness or contentment, and very rarely described their quality of life in terms of pain or distress. Instead, they very much focused on their ability to live a 'normal' life, on attending school, taking exams, doing things with family and friends, engaging in meaningful activities, and gaining independence from their family.

When discussing their need for further treatment or medical intervention, young people often described their symptoms in this way, drawing on the medical and physical indicators but within a narrative of their everyday lives.

"When I was at school I used to fall asleep a lot, about half two when I was at school, in lessons. And I was waking up with headaches in the morning. It was like having a hangover every morning, not nice."

Harry talks about the impact of his respiratory problems on his quality of life.

For Harry, having continuous chest infections meant that he spent weeks on end in bed, therefore missing school and his life in general. It also meant spending time in hospital, which was not something he enjoyed. Therefore the wider impact of having chest infections seriously impeded his quality of life during the weeks in which he was unwell.

“I’ve always been in and out of hospital with chest infections.....If I’d get a cold, I used to be in bed for like three weeks, and not really eating anything, just drinking, and then I used to lose loads of weight.....”

Harry talks about the loss of weeks at a time when he became unwell.

Richard and Simon too focused on the wider impact of the symptoms they experienced on their everyday life, having both been recommended to have a gastrostomy. Richard, in considering the impact of aspiration, drew attention to the lengthy stays in hospital and the disruption they caused in his life, and this information was used in forming an opinion that he might benefit from a gastrostomy. In contrast Simon, who also talked about assessing his quality of life at the time it was recommended, believed that although he was underweight and found it difficult to eat certain foods, it had little impact on his life or wellbeing, and because of this he decided against it.

However, like parents, young people can find themselves in conflict over the different elements that make up quality of life. For both Alex and Matthew, this compromised their ability to make a decision about technological support because they both felt that their quality of life, despite their deteriorating health, was at the time of making the decision very good.

“That was an interesting one that was coz I was in between, I didn’t really know.....it would keep me well and stuff so I could keep doing things. But at the same time I was going out into the world at the time. I just thought, “do I do the PEG or not?” You know, I didn’t really understand why?”

Alex admits not being able to understand why he needed a gastrostomy.

“At the time I decided well, I felt like I’ve got more quality of life at the moment than to do it.”

Matthew felt that his quality of life at the time of making a decision was good.

The conflicting elements that make up quality of life are crucial to understanding the dynamics of decision-making for children with life-limiting conditions, because they provide an insight into the risks parents and young people are willing to take in order to maintain or enhance quality of life when it is known a child or young person’s life is compromised. These dynamics are not limited to decisions about technological support, and are integral to many of the decisions parents and young people make regarding their lives.

“She wanted to parachute jump.....well my friend said “how can you let her go, what if anything happens”? And to be honest all I could think was, “well what a way to go for her. Doing something she wants.” ”

Kate’s mum on taking risks in order for Kate to achieve her dreams before she died.

“We have a caravan on the coast, and he used to disappear off, all round the caravan site, on his own, which is what they want, they want to be independent.....You’ve got to let them do it, and you know when he was gone for ages you used to think “oh my god where is he? What’s happening?” ”

Peter’s mum used to worry about the risk to Peter being out on his own.

“I eat crisps, and sausages and chips. All sorts of things like that. They all look at me like, you shouldn’t be eating things like that. But I don’t care.”

Alex talks about his quality of life, and the risks that are worth taking.

7.1.3 Child’s quality of life so far

Whereas for young people the focus is very much on their quality of life in the present, and in the aspirations and goals they wish to achieve, for parents quality of life is both complex and fluid, and parents can find themselves continually re-adjusting their expectations for their child, and their understanding of their child’s

quality of life, particularly in the case of children whose conditions are degenerative or progressive.

Dad: "I mean he likes the play station but he struggles using that now, so it's usually pictures and shopping. But he's content just watching someone else playing the play station."

Mum: "As long as he's in with, you know, he likes to be in among it rather than on his own doesn't he?"

Sam's parents describe re-adjusting their expectations regarding his quality of life, and finding new ways in which Sam can find meaning in his life.

For some parents, the deterioration in their child's quality of life over time, and the suffering they have experienced from their symptoms, and as a result of the treatments and interventions they have received, lead parents to believe that further intervention or surgery is not appropriate.

"We went through so much with Josh when he was born and when he was in PICU. You know I got to a point where I thought "no more, just leave him alone". You know, no more."

Josh's mum takes into account how much Josh has already suffered.

Following Sam's gastrostomy and spinal rods, his parents have now decided that Sam has endured enough, having spent a great deal of time in hospital due to the complications arising from these procedures, and consequently missing out on school and other activities he enjoys.

Mum: "We've made up our minds now."

Dad: "You know, to the best of our knowledge he's not having any more."

Mum: "No that's it now, unless it's you know his appendix or something that can't be helped".

Sam's parents describe their decision for Sam to have no further interventions.

Lewis's parents expressed their final decision for Lewis regarding technological support in this way, a decision that was made at the end of life, and one that was based on both Lewis's quality of life at the time of making a decision, and on the life he had already lived.

Mum: "Our decision was based on what Doctor [Name] had said, but also based on his entire life and all his struggles, and everything we'd watched him go through, you know. If you'd taken that in isolation and said "make this decision". If you do this, this could happen. If you do this, this could happen. You might make a different decision, do you know what I mean?"

Dad: "And we thought, you know, "he's been through enough now". You know what I mean, it's a case of "no, we don't want any more surgery to put more tubes in him or anything like that"."

Lewis's parents talks about the importance of the life Lewis had already lived.

Distinguishing between the quality of life a child has already lived, and the quality of life a child is able to achieve at the time of making a decision is significant, and for parents who describe making their decision in this way the option of doing nothing becomes a real one to consider. However, some parents can find this difficult, particularly when it is felt that a child will not survive without the initiation of technological support. In these cases, the decision is about the need to sustain life whatever the cost, and for parents the emotional response to save their child's life when it is acutely threatened is almost impossible to overcome.

"You've got to kind of say "well yes", because you clutch at anything don't you, to keep them alive. You know, you just clutch at anything they offer you that might just prolong their life, that bit longer you know."

Peter's mum talked about the difficulty of considering doing nothing as an option.

Nonetheless, for decisions about technological support the temporal dimension to quality of life is important because two children who appear to have a similar

quality of life at the point of making a decision may well have experienced life up to that point in very different ways. Combining the knowledge of their child's current quality of life, and the life they have already lived therefore enables parents to consider whether the additional burden of initiating technological support is worthwhile.

Mum: "We're looking at her quality of life and so if anything major was to happen, I mean obviously you know we would try and, if she stopped breathing or whatever we would try and do, but we wouldn't go for full jumping up and down on her chest."

Dad: "But if her progress was developmental we wouldn't have that."

Mum: "Yeah, if she was, even if it was very slowly, but she was developing, we wouldn't be making these decisions. But it's because she isn't, and if anything has regressed."

Hollie's parents discuss Hollie's cognitive deterioration over time.

7.2 Weighing up the Potential Outcomes of Technological Support

Whereas the preceding factors are used in assessing if there is a need for medical intervention, the second set of decision factors are used in considering the range of potential outcomes associated with the intervention being recommended, which are then used to weigh up the benefits and drawbacks for the individual child. However, there is considerable variation in the chronology of this process, despite the logical order implied. Whereas for some families, considering the potential outcomes does come after assessing the impact of a child's difficulties on their quality of life, others find themselves weighing up the potential outcomes of a gastrostomy or ventilation having been previously unaware that their child has a need to be considered for technological support.

In both decision scenarios the two components of the decision process are not mutually exclusive and are intertwined within the accounts of parents and young people. Richard's decision about a gastrostomy illustrates the way in which families

move back and forward between the two in order to make a decision, as being able to understand the impact that aspiration had on his quality of life helped Richard to identify the potential benefits a gastrostomy might bring, a technology that he had been very reluctant to consider at first.

“I’d been in hospital with a chest infection about three times in the space of two months.....she said that this would help fight off infection and be stronger, and then I wouldn’t have to come into hospital so much.”

Richard identifies the impact of chest infections on his life.

Nevertheless, distinguishing between the two components of the decision process is important because for some families a decision has already been reached by the time a recommendation is made for technological support. This decision is not necessarily about the intervention itself, with some families forming an opinion that their child requires medical intervention, and other families coming to the opinion that in fact the quality of life their child can expect to achieve may not warrant further action. Ultimately, believing their child needs medical intervention helps parents to accept the risks and uncertainty involved in agreeing to a gastrostomy or ventilation, and it is therefore a crucial element of the decision process.

The relationship between the two components of the decision process also illustrate the complexity of perceived choice in decision-making, because in essence families are describing a process in which by considering whether or not the impact of their child’s condition on their quality of life requires medical intervention, they are in fact considering the choice of doing nothing. However, only some of the parents and young people viewed the process in this way.

“With the trachee it was quite straight forward.....there was two choices, one you just do nothing and it would be the same as I was. So nothing was changing. So you either do nothing and nothing changes, or you do something and it does change.....And I thought, why would I want to

spend three weeks in bed, not eating [his life before the tracheostomy], why would I want to do that? I don't have to. Why would anybody choose to do that?"

Harry talks about a decision of two choices.

7.2.1 Risks and complications

Regardless of the opinion with which parents arrive at the recommendation or suggestion, both the risks associated with inserting a gastrostomy or initiating ventilation, and the complications that may arise once a child or young person begins feeding by gastrostomy or using a ventilator, are key decision factors. These risks are central to the decisions parents and young people make, and they are sometimes able to recall with clarity a single risk that has influenced their decision.

"She would need to go to Intensive Care afterwards and all the you know potential problems for it, and that there are 10% of the kids who have it done who still suffer the regurg, who don't you know, even with it, it's still a problem.....plus then you're at constant risk of infection by having you know having it there."

Hollie's mum describes the risks and potential complications of a gastrostomy.

For others families, the actual risk is difficult to predict due to the complexity or uncertainty regarding a child's condition.

"If they go in and miss it, even the most skilled person, they can perforate the bowel. They've got to, you know, he's all concaved, his chest, and he's bent over, his hips are funny.....Well they'll know, he says "it's a risk that it could not go right", and then going under, well last time he was under there was a hell of a job getting him out."

Steven's dad revealed the uncertainty regarding a gastrostomy.

One of the central risks parents discuss regarding a gastrostomy, and also a tracheostomy, is the risk of surgery and the associated risk of general anaesthesia. For some children, this risk is described as significant due to respiratory or other weaknesses caused by their illness.

“We were very worried that Sebastian was so frail that he might not survive the operation. When it was decided that he should have the operation, and we went and saw the surgeon and the surgeon said “well because of his condition you realise it’s a 50:50 chance of him coming through it. It may not be successful.” That was an enormous strain.”
Sebastian’s dad recalled the level of risk involved in a gastrostomy.

The risk that their child might die during surgery is one that parents find emotionally difficult to weigh up, and in Alex’s case, while the gastrostomy itself was not considered by Alex or his mum as an end of life decision, the risk to Alex of having general surgery led Alex’s mum to make the decision on this basis.

“My son’s never gonna die on an operating theatre. He hasn’t come this far to die on one.....And each time I’ve taken him down for surgery or for one thing and another I’ve had to say goodbye because you never know if he’s gonna come back out.....I mean how many more times do I have to say goodbye?”

Alex’s mum describes the end of life nature of decisions requiring general anaesthesia.

The hospital stay itself is regarded as a potential risk, with parents referring to the variation in standards of care, the difficulties and mistakes they have encountered during previous hospital stays, and the lack of specific knowledge about their child’s condition they report some of the ward staff to have. Furthermore, parents identify the higher risk of infection or illness during periods of hospitalization, which is a specific concern when their child is medically vulnerable due to their nutritional or respiratory difficulties.

“They gave him this operation. Then they stick him in a ward with three other people all with chest infections. Oh good! So then he gets a chest infection.”

Harry’s dad emphasises the need to keep Harry’s hospital stays to a minimum.

Assessing the risks and complications associated with the administration of technological support is central to decisions. However, the process of doing so is

identified as a challenge by parents due to the complexity of their child's condition, and the uncertainty surrounding the risks identified. What's more, parents consistently report feelings of fear, worry, and doubt when weighing up the risks and benefits for their child. Young people also find the risks difficult to accept and recall the negative feelings that accompanied the process of decision-making, even when the potential benefits were easy to identify.

"I've always been quite strong minded in terms of wanting procedures to benefit me, and realising that, it felt like it was sickening at heart to do it, but I had to have these procedures."

Richard recalls his initial feelings about having a gastrostomy.

In attempting to understand the risks, some families draw on their knowledge of other children with a gastrostomy or ventilation, who may or may not have experienced difficulties or complications. Parents and young people sometimes report using this information to form an opinion about the possible outcomes, therefore influencing the decision they ultimately make. In some cases, the salience of a particular complication experienced by someone known to them becomes central to their decision, and is not necessarily based on the actual level of risk involved, which may be lower than for other complications.

"But you have problems, all his friends with buttons.....you do get infections."

Steven's dad recalled his knowledge of other children with a gastrostomy.

Parents and young people also report drawing on their own experiences, comparing the risks of a gastrostomy or ventilation to those of other procedures they have already considered. In weighing up the level of risk involved in having a gastrostomy, Richard drew upon his experience of having spinal rods, a procedure he believed was much more complex. This comparison enabled Richard to accept the lower risk involved in having a gastrostomy.

"I'd had spinal surgery by then so I knew kind of the risks of that and I knew that I'd come through that OK.....and also I think because I'd experienced a procedure, such a big one with my spine, that it was maybe not even, I felt that any operation was small compared to that."

Richard compares the risks of a gastrostomy to those of having spinal surgery.

Matthew's mum too spoke of comparing the risks of assisted ventilation to those of spinal rods, the latter of which Matthew had previously decided not to have.

"The risks involved, and I mean, at the end of the day, well if it were gonna be a cure I'd have it done but it's not. It might enhance my life, it might not.....But I think this was something different because there were no risk involved with the ventilation where the spinal operation there were lots of risks involved, just with the operation."

Matthew's mum compared the risks of ventilation to those of having spinal surgery.

7.2.2 Invasiveness

In fact, although the study selected two examples of technological support to explore, it was evident in the data that families viewed the risks of initiating non-invasive ventilation as less significant than those of having a gastrostomy or a tracheostomy. Similarly, a gastrostomy was compared to spinal rods and other surgical procedures their child had received in their attempts to assess the risks of the intervention being recommended. This aspect of the decision process very much relates to the invasive nature of the intervention being considered, which was identified as an important factor in the decision being made.

However, invasiveness as a decision factor is multi-dimensional, and takes on more than one meaning for families as they weigh up the potential benefits and drawbacks of the proposed technology. Firstly, the need for an intervention that requires the surgical insertion of a foreign object within the body is a consideration for parents and young people, and one that is viewed separately to the risks associated with the surgery itself.

"It was the fact the gastrostomy was physically in Casie.....That wasn't how I'd given birth to her and it was, it was the kind of physical sight of it being there I couldn't handle."

Casie's mum found it difficult to accept the gastrostomy due to its invasiveness.

"I mean I remember going to see somebody who had a tracheostomy and it's just hard, it was hard really, seeing this hole in their neck.....so I was quite taken back by that when I saw a child with tracheostomy. It's quite a big shock."

Josh's mum describes her reaction to the invasiveness of a tracheostomy.

Secondly, the invasiveness of the actual technology and equipment required is something parents and young people include within the process of weighing up the benefits and drawbacks. For example, many parents regard the nasal gastric tube as more invasive than a gastrostomy due to where it is positioned and the impact it can have on a child when it has to be re-inserted. Charlotte's mum, for example, remains convinced that her daughter's refusal to eat, which in part led to the recommendation for a gastrostomy, was caused by the invasiveness of an NG tube.

The invasiveness of ventilation is also assessed in this way, with some young people like Matthew, who made a decision about non-invasive assisted ventilation, identifying the invasive nature of the mask as a decision factor. Alex and Harry, who now have a tracheostomy but who had in the past used assisted ventilation of the 'non-invasive' type, describe the mask as invasive, more so, they believe, than a tracheostomy.

Richard too refers to invasiveness in this way, comparing a medically invasive gastrostomy with non-invasive assisted ventilation.

"I think with the gastrostomy you don't actually feel it because it's, well because like, it's hard to explain really, even though it's in your body and it's in your skin you can't feel it. It just feels like your stomach. Whereas when the mask is on, it's pressing on your skin, and so sometimes I used

to get like breakages of skin and sores and stuff.”

Richard compares the invasiveness of a gastrostomy to ventilation.

Finally, some parents and young people spoke of invasive in aesthetic or visual terms. For example, Matthew felt that the presence of a ventilator and mask would make him appear more disabled, and Harry’s dad referred to the mask in terms of how it might appear to other people.

“It’s just another physical thing that shows me disability if that makes sense.....slightly embarrassed having it on and stuff, people seeing me with, being attached to a machine.”

Matthew worried that ventilation would be aesthetically invasive.

“And the people who are wearing masks of course, I mean we saw one a few weeks ago....And we saw this lad with this mask on, he looked like a dalek.....And why? Why?”

Harry’s dad talked about the aesthetic invasiveness of a mask.

The multi-dimensional nature of invasiveness is important, and it cannot be assumed that when a family deliberates over how invasive a procedure might be, they are referring to invasive in medical terms. Furthermore, some families may consider several dimensions of invasiveness when considering the benefits and drawbacks of a gastrostomy or ventilation. For example, Harry and his parents identified the physical invasiveness of the ventilation mask as a key driver in the decision they made to pursue a tracheostomy, against the recommendation of Harry’s consultant to continue using a mask. Yet at the same time they still described the tracheostomy as both medically and aesthetically invasive, and included this within the decision about whether it was appropriate.

“The trachee doesn’t look particularly attractive and I think that puts them off a little bit. But in terms of what he can do, where he can go, and the freedom it gives him, and the lack of potential injury to his, you know, to his wellbeing.”

Harry's dad identifies the different meanings of invasiveness with regards to a tracheostomy.

"It's a strange thing, when you think of a tracheostomy you think of it as a, quite a major thing to put a hole in your neck. It's not like it's something you see every day."

Harry recalls the surgical invasiveness of having a tracheostomy.

7.2.3 Permanence

Like invasiveness, permanence is multi-dimensional, related to the long-term impact that the proposed technology will impose on a child's life, and the progression in a child's condition that is brought into focus when decisions about technological support are made. Permanence is also assessed in terms of the reversible nature of the decision outcomes, and in considering the risks of a tracheostomy Harry recalls being reassured that if the outcomes were poor, the tracheostomy would heal up and he could resume ventilation by mask.

"Because with a tracheostomy it can be like, can be sealed over again, then it's not like a permanent thing.....once you've done it, it can grow back. But with some things you can't obviously change your mind once you've done it, but with that it's not actually a case of if it doesn't work you're stuck with it. You can go back to how you used to be. When I decided to do it I wasn't thinking that then. I was thinking "yeah this is the right thing, it's gonna work, it's gonna be brilliant". But obviously in the back of your head there's always that thing that you can if you need to, get rid of it. That it's not a permanent fixture, kind of thing."

Harry talks about the permanence of a tracheostomy.

Although Harry describes not explicitly using this information to form an opinion, it did help to reduce the anxiety he experienced regarding the uncertainty about whether or not a tracheostomy would benefit him. Harry's dad too refers to the permanence of a tracheostomy as a decision factor.

“Even if it didn’t work we could just close it back up again.....we could just take it out and it would just close up. And then he’d be back to square one where he was before it was suggested.”

Harry’s dad recalls discussing the reversibility of a tracheostomy with Harry.

Other young people and parents discussed the perceived ‘permanence’ of a gastrostomy or ventilation, and again the reversible nature of the intervention was used by families to manage the uncertainty associated with weighing up the risks involved, and the difficulties in assessing the potential impact of the technology on a child’s life.

“A PEG I think was originally, they thought he might grow out of it but they realized he wasn’t going to grow out of it, you know the condition.....And they always said to us that if it ever came out that it would heal up. Because I was worried that you know would it heal back up. And they said “oh it wouldn’t be a problem”. So I think realizing that it would go if it needed taking back out, it would go back to normal and it would be fine sort of thing, just a scar. And that would be it really so, it wasn’t like a big major decision.”

Josh’s mum recalls making a decision on the basis that Josh may not need a gastrostomy permanently.

The passage above also illustrates the uncertainty parents can experience about whether or not the technology being recommended will be needed on a long-term basis, and this meaning of permanence was a factor in Matthew’s decision about assisted ventilation, particularly as the recommendation came at a time when Matthew was still able to manage sufficiently without assistance from a ventilator.

“I mean that was some of the questions Matthew said, you know, do I have to be on it? And once I go on it, is that it, I can never come off. And obviously that wasn’t the case.....”

Matthew’s mum highlights the uncertainty about the long-term nature of ventilation.

In stark contrast, the decisions Alex and Peter made regarding assisted ventilation were due to the life threatening situations they found themselves in at the time it was recommended. Without ventilation, it was felt that neither would survive and the decision was therefore made with the knowledge that ventilation would become a permanent feature in Alex and Peter's lives.

"He knew it would become a permanent feature in his life. It wasn't going to be, you know, we couldn't change our minds next week and not bother. We go along this path or we don't? You can't use it two days and then not use it three days. It's every day."

Peter's mum on the permanence of ventilation.

The meaning of permanence for young people who are considered for assisted ventilation is not straight forward, particularly for young people who begin using a ventilator at a stage when they can still breathe unassisted, who sometimes only recognise the long-term nature of the decision they have made as they become increasingly dependent on the ventilator. In those cases where the decision being made is about the use of overnight ventilation, young people are not always aware that the ventilator might over time become a permanent feature in their life as their dependence increases and they move from only needing assistance overnight to having to rely on it during the day as well.

Matthew's mum in fact views these decisions as separate due to the potential impact of being permanently ventilated on Matthew's quality of life. Harry also talks about the difference, although admits that once on overnight ventilation the move to full ventilation appeared to just happen without a conscious decision being made about the implications this might have.

"I mean I do know that Matthew's already said he wouldn't want to be on ventilation day and night. He sees that as not a good quality of life, and we've talked about already about things like life support and things like that, so we know."

Matthew's mum distinguishes between overnight and full ventilation.

The added complexity of permanence raises concerns about the informed basis upon which decisions regarding technological support are made, and highlights the potential conflict in making a decision about a technology that is sometimes viewed as reversible by parents and young people, despite the permanent feature it will become in a child's life and the impact it will therefore have over the long-term.

7.2.4 Quality of life

Quality of life is central to the process of weighing up the potential impact of technological support, and both the quality of life at the time of making a decision, and the life a child has already lived become an important reference point when families consider what the potential benefits and drawbacks of a gastrostomy or ventilation might be. The data revealed a range of benefits parents and young people consider, and while diverse because they concern the life of the individual child or young person, the decisions are ultimately about minimising the impact of a child's condition on their quality of life, as the following passage illustrates.

"I think sometimes people have got to realise that your decision will quite often be about your life and about the benefits that it can make to allow you to live a more independent life and to be able to do more things. It's not just for the medical side so where a lot of doctors and staff may think oh well it improves the medical side so they should come and stay in hospital for a year or whatever, a long period of time, and have this operation, which is really minor and doesn't really improve your medical side that much, but by doing it you'd be missing out on so much. I think that's quite important. Make sure they understand that people have got to live their life even though they have a condition. That's very important really."

Richard sums up his approach to considering the benefits of a proposed intervention.

Viewed in this way, the health-related benefits form only one element of an overall assessment of the potential impact technological support will have. Nonetheless, it

can be difficult for parents and young people to weigh up the potential benefits of an unknown procedure, particularly one that is accompanied by impacts that are less positive and easier to identify, such as the potential discomfort and invasiveness of living with technological support.

“Having something stuck on your head all night and being told that I had to kind of, essential to wear it and you think “well it doesn’t seem to do anything”especially as a child it’s hard to understand the difference.”
Richard found it hard to identify the benefits of assisted ventilation at the time it was suggested.

The hospital stay was identified by families as a potential drawback, and although it is a short-term one, the importance of this factor in the decision was evident in the data, especially in the accounts of young people. In contrast to parents, who focused on the risk to their child’s health of staying in hospital, young people drew attention to the boredom and isolation during hospital stays, and the unpleasant environment and food. Where the initiation of technological support requires a long hospital stay young people consider this in the decision they make, and Simon chose not to have spinal rods due to the length of time he would have to spend in hospital. Matthew also identified this as part of the decision process, and being able to begin ventilation at the children’s hospice where he stayed for regular respite was for him a key decision factor.

“I went to [children’s hospice] to actually use it for the first time.....coz they’re used to all the overnight ventilation, the staff there. So they know what to expect and stuff.....and because it wasn’t a hospital so that’s a plus sign, and also because they were people that I already know.”

Matthew reveals the benefits of beginning ventilation in an appropriate setting.

It is not surprising then that for young people one of the key benefits of technological support is the thought of not having to spend as much time in hospital as a result of the symptoms and difficulties they have been experiencing. Indeed for

Harry, although ventilation would alleviate his respiratory problems and reduce the risk of chest infections, it was the idea of spending less time in bed and in hospital that led to the decision to proceed with a tracheostomy, despite its invasiveness and the additional care a tracheostomy would require.

“Without the trachee it would be the same as I was. So nothing was changing.....And I thought “why would I want to spend three weeks in bed, not eating, why would I want to do that? I don’t have to.”

Harry identified the key benefit of ventilation as spending less time in bed.

Richard also identified less hospital admissions as a key benefit of both ventilation and a gastrostomy, and one that helped him make decisions about them.

“She [Richard’s paediatrician] thought that I wasn’t eating much and this was partly why I was coming in with chest infections. She said that this would help fight off infection and I’d be stronger, and then I wouldn’t have to come into hospital as much. And you know that was a positive thing for me. And I could understand that.”

Richard identifies less hospital admissions as a benefit of a gastrostomy.

For Steven’s parents, the experience of his NG tube enabled them to identify the potential impact of a gastrostomy. This led them to believe that the benefits of artificial nutrition in general were not enough to justify a gastrostomy, particularly considering Steven’s love of food, and the ease with which his parents felt able to feed him. The advice they had been given that Steven should not gain too much weight also led them to question the benefits of a gastrostomy for Steven. Like other parents, their encounters with children similar to Steven who were fed by gastrostomy provided them with an insight into what life might be like for Steven if he gained weight.

“He’s always been on the thin side but he’s always ate right.....So what we were saying about weight, they were trying to pile weight on him.....And she took us down to see these boys, and they were like two

pork pigs, and I thought I don't want Steven like that. And they were just laid there. What life is that, it's not a life I would want.....You see we take him all over the place."

Steven's mum talks about the quality of life he might have with a gastrostomy.

On reflection, Steven's dad, who is Steven's primary carer, was able to identify the benefits of providing fluid and medication by gastrostomy, but this did not form part of the decision at the time. This was in part because the recommendation was based on a clinical diagnosis of aspiration, and on the need for Steven to gain weight. The wider benefits of a gastrostomy were therefore hidden, and in hindsight Steven's dad believes that a gastrostomy could have improved Steven's quality of life due to the distress caused by administering his daily medication, and the on-going problems with constipation due to suboptimal fluid intake.

"It's hard work with all them drugs. Now then you see if there's a gastrostomy, there's no problem, it goes straight in you see."

Steven's dad talks about the benefits of a gastrostomy for Steven.

In contrast, Emily's parents were able to identify the wider range of benefits associated with artificial nutrition because of the immediate and positive impact an NG tube had on Emily's life.

Mum: "Within weeks she started to put weight on"

Dad: "Ahh, she were different as well because she were getting her medication wasn't she?"

Mum: "Yeah.....sort of her fits cut down by half, and she wasn't hungry. So she didn't have belly ache."

Emily's parents identify the benefits of artificial nutrition for Emily.

Kate too considered the potential benefits of having a gastrostomy in terms of the impact on her life as a whole. Although she made the decision before tube feeding was recommended, Kate was aware that without it she might experience greater distress at the end of life because of the severity of her symptoms and pain at this

time. However, having lost a close family member to the same progressive illness, Kate believed that her quality of life would be so minimal at the stage a gastrostomy might be recommended, that it would only prolong her suffering at the end of life.

“She couldn’t swallow, she couldn’t speak, and before it became this bad we sort of talked about it, you know “what do we do?” And we decided, well she decided, with my agreement, that when the time was right she would want to stop her medication which would mean that would be the end after a little while. So that was the decision we had to make.”

Kate’s mum explained the rationale behind Kate’s decision not to be tube fed or medicated.

7.2.5 Loss

Decisions about technologies that are life-sustaining can bring into focus the progression of a child’s illness, and force parents to think about and re-adjust their expectations for their child’s quality of life as they face the potential loss of their child’s ability to feed or breathe independently. Decisions of this nature can also create conflict between the different elements that make up quality of life, with a gastrostomy or ventilation in some cases improving one aspect of quality of life while at the same time adversely affecting another. In some instances, this can create a sense of impending loss, which becomes a factor in the decision families make.

“I couldn’t handle it at all. I just couldn’t handle it. It was, for me, losing the ability to feed Casie normally was such a fundamental thing.....because to me, I needed to be able to feed her.”

Casie’s mum describes the emotional attachment she had to feeding Casie.

With regards to a gastrostomy, the sense of loss is about more than the loss of a bodily function and can be highly emotive for parents, who express a range of feelings about the potential loss of a parental role. Some parents report feelings of guilt from having to acknowledge that they are not able to provide their child with adequate nutrition. Others describe the sadness and sorrow they feel for their child,

who they believe will lose an enjoyable and social aspect of their life. In fact, for some parents the decision is not about the gastrostomy. It is, in essence, about the psychosocial and emotional impact of agreeing to feed their child artificially, and what this means for their child, for themselves as primary carers of their child, and for the family unit as a whole.

“What am I gonna do with her now? You know, 11 o’clock we’d put songs on, give her a yoghurt just to keep her going. That were lost. Plus I used to think “what can I feed her that’s fattening, mashed potatoes and fish in butter sauce”. I’d be mashing it all up, one for the freezer, one for today, one for tomorrow. And all that stopped. So half me day, half me jobs had gone because she didn’t need me to mash her food up, or to feed her.”

Emily’s mum recalls the loss of interaction and bonding, of parental duties.

In some cases, the perceived reversibility of a gastrostomy or ventilation helps parents accept the potential loss that can be associated with a decision. For Charlotte’s parents the anticipated loss of oral feeding was a key decision factor, and at the time a gastrostomy was recommended Charlotte’s mum felt that giving in to the gastrostomy was like giving up on the idea that Charlotte would eat again. The reassurance offered by the consultant that Charlotte might feed independently in the future, and that the gastrostomy could be reversed if this happened, alleviated the sense of loss and enabled Charlotte’s mum to agree to a gastrostomy.

This contrasts starkly with the decision Robert’s family made about a gastrostomy. Robert’s mum describes succinctly the reasons for Robert’s gastrostomy, to ensure he received adequate nutrition, to reduce the risk of aspiration, and to reduce the burden on Robert, and the whole family, of having to spend hours each day feeding him. As Robert’s mum points out, the benefits more than outweighed the risks, and because Robert could continue feeding orally following his gastrostomy, he was still able to take pleasure from this activity, and enjoy mealtimes with his family.

Decisions about ventilation are also associated with a sense of loss. Josh's mum spoke emotionally about the anticipated loss she experienced regarding her child's voice, which would be temporarily lost as the result of a tracheostomy. Josh's mum explains that these feelings were intensified because of the other limitations her son would experience due to his condition, and the sense of loss she felt as she came to terms with his diagnosis.

"I remember when they were telling me about that, what the tracheostomy was, and that, and how he wouldn't be able to talk. That really hit me. That really hit me. It was hard.....You know they said "oh you wouldn't be able to hear him talk".....that took me a long time to come to accept.....coz I thought oh you know, there's having a sick child and then knowing that you're not even going to hear his voice sort of thing. That really sort of got to me."

Josh's mum describes how upsetting it was that Josh would not be able to talk following the tracheostomy.

Josh's mum admits that agreeing to Josh's tracheostomy took a long time, during which she swayed between the two positions. In doing so, Josh's mum, like other parents, drew upon the knowledge about why Josh needed a tracheostomy and what it would mean for his life in order to come to terms with the feelings of loss she experienced.

Where young people require technological support, decisions can be about the loss of independence caused by the need to rely on equipment and machinery, and both Matthew and Harry talked about the need to have a carer present more often because of their ventilation.

"I need a carer there nearly all the time because if it gets blocked or anything like that, because they need to sort it out, but that's the only issue"

Harry identifies the loss of independence from having a carer present at all times.

It can also be about the loss of normality, as the ventilation is yet another indicator of a young person's limitations and disability. Alex's mum talked about the loss of autonomy, and for Alex, the loss of oral feeding was associated with losing one of the few things in his life over which he had control.

"They thought it would be easier to just, "well can't you eat anything healthy". Well, Alex looked at and went, "well go on it's not as if my body's healthy is it?" And at the end of the day he enjoyed a burger and chips. "And if that's all I have in life to enjoy then I'm gonna eat a McDonalds".....But then that was his choice. He felt, that was good, because at least he has a choice on what he can eat."

Alex's mum explains that the loss of food was also about the loss of independence.

Sometimes the need for ventilation follows a life-threatening event. In these cases the loss of life becomes the central concern and it can be difficult for parents and young people to consider other quality of life factors in this instance.

"Actually, I thought, I don't like this so I was a bit hesitant obviously, I mean I didn't want to do it.....But I could hardly breathe, I had to just hit and miss with it. Do you know what I'm saying? Coz if I hadn't I'd be dead. I just had to do it, just hit and miss, otherwise I would have died.....it was either trachee or die."

Alex describes the end of life nature of his decision regarding a tracheostomy.

"You think is it worth all the hassle and then you think, well yes because I don't want to lose him.....It didn't prolong his life that much more, about a year, but you don't know that at the time do you?"

Peter's mum recalls the life or death nature of the decision her and Peter made.

7.3 Chapter Summary

The decision factors presented in this chapter relate specifically to the decision under consideration, which is distinct to the wider context and process around it. Providing a closer look at each factor in turn again illustrates the central themes of

uncertainty and perceived choice arising from the findings of this work. Chapter Eight will now examine the category of decision features, in other words those features about the patient population identified as having the potential to influence the decision process, and ultimately the perceived choice experienced by families as they weigh up the appropriateness of technological support.

- CHAPTER 8 -

DECISION FEATURES

This chapter provides a detailed account of the category of decision features shown in the model presented in Chapter Six, in other words the features of the patient population, in this case children and young people with life-limiting conditions, identified from the research that influence the decision process. The chapter contains five key themes beginning with a discussion of how the prognostic uncertainty associated with many life-limiting and progressive conditions can influence the way in which families make decisions. This is followed with an account of how the individual child can influence the process of decision-making, and their developing role in making decisions about their condition. Next the evolving role of parents as decision-makers is discussed, and the chapter finishes with a consideration of how beliefs and values can influence the process of decision-making, and the role of gut instincts and intuition.

8.1 Prognostic Uncertainty

The prognostic uncertainty associated with childhood life-limiting conditions is identified from the data as a key feature in the decisions families make regarding technological support. As parents and young people recalled other decisions they had made about the management and treatment of their condition, it also became evident that the impact of prognostic uncertainty both varies across families, and across condition types. Despite this diversity, prognostic uncertainty is an underlying feature in the lives of children with life-limiting conditions, and therefore presents a challenge for families as they attempt to weigh up the potential impact of a proposed intervention which is already surrounded by uncertainty due to the combination of risks, complications, and benefits.

Chapter Seven identified the stark contrast between making decisions for children with well understood conditions, compared to those with undiagnosed or rare

illnesses about which little is known, and this contrast is related to the varying uncertainty surrounding the very different prognoses. For children who have an undiagnosed or very complex condition, or for when a decision about technological support comes before a diagnosis is made, assessing a child's needs can be difficult because parents experience uncertainty about whether the need relates to the progression of their child's condition, or to a separate undiagnosed, or unrelated problem. It can also be difficult in these situations to determine if another solution is available when there is such uncertainty about the cause of the symptoms leading to a recommendation for technological support.

Charlotte's Gastrostomy – Case Study

Charlotte, who has very complex health care needs caused by a combination of related conditions beginning at birth, was able to feed well for the first two years of her life. However, she began to experience painful and distressing symptoms of reflux that due to the complexity of her condition went untreated for a year. By the time her symptoms subsided as the result of successful treatment for another medical problem, Charlotte had developed a phobia around the mouth area and refused to eat or drink, or to take medication.

At the time, Charlotte's parents felt it was important that something be done due to the risk of malnutrition and Charlotte's underweight state. However, her condition was not well understood and the reasons for her feeding difficulties remained unknown. Despite this, and even at the insistence of her parents to try alternative solutions to encourage Charlotte to feed orally, the consultant involved in her care insisted that a gastrostomy was the best course of action, and Charlotte's parents describe feeling pressured to agree.

Although they believe that in the short term a gastrostomy was life-saving for Charlotte, they express continued regret that over the long term it was not in Charlotte's best interests due to the uncertainty about why Charlotte needed artificial nutrition at the time and the lack of understanding about her overall condition.

Charlotte's case illustrates the desire parents have when making a decision to understand why their child needs technological support, and the range of emotions parents can experience when there is uncertainty regarding their child's condition or about the reasons for their child's symptoms. In Charlotte's case, her parents

continue to experience regret about the decision they made, and reflecting back on the decision was very upsetting for Charlotte's mum, despite the positive outcomes for Charlotte following her gastrostomy.

Lewis's parents also recall finding it difficult to make a decision due to the prognostic uncertainty at the time, which continued throughout Lewis's short life because neither a diagnosis nor a realistic prognosis was available. Having to decide whether or not to withdraw ventilation after Lewis was born was therefore very difficult because his parents were unable to make a judgement about what sort of life Lewis would have if they chose to ventilate, or whether in fact he would survive at all if they chose to withdraw.

Mum: "It was a bit, harder and harder to know what the future was really because we didn't know."

Dad: "We didn't really quite know how severe it was did we?"

Mum: "No I mean it was all new to us you know, we were thinking "well crikey, well, put him back on again you know, kind of thing". Well you know, very difficult decision.....We needed to know more didn't we?"

Lewis's parents describe the prognostic uncertainty around his condition.

Lewis's parents reflected back on the information they were given about his prognosis, and the uncertainty expressed by the consultant looking after Lewis at the time of making a decision.

Dad: "That first decision was hard, because they couldn't really say at that point how things were.....When I asked something specific about you know "how long is he going to live?", "what's he going to be like?" I mean, and he just kind of went [shrugs shoulders], shrugged his shoulders."

Mum: ""The bad end of bad" he said. That's what his phrase was. "It's the bad end of bad." "

Dad: "Yeah"

Mum: "That was his words. And you're just like, well you don't want to hear that do you, do you know what I mean? But I mean he was right. He was absolutely spot on, but, and then when I said to him about his life expectancy he said "well look you know, because he's so, he's physically, perfectly formed, you know he could be sort of end up being as strong as an ox and outlive you or he could succumb to, you know, an infection", as he did in the end."

Lewis's parents describe the prognostic uncertainty expressed by their consultant.

Lewis's parents described the range of emotions they experienced during this time due to the prognostic uncertainty around the decision process, and this was also identified by Emily's parents and Josh's mum.

"We didn't know a thing. Because she didn't have a label you see so, that were rough wasn't it coz like we didn't know what were happening.....so it were like very scary at that time."

Emily's mum recalls not knowing anything about Emily's condition.

"Well that wasn't the underlying problem. At that time he hadn't been diagnosed for what his condition was. But they knew he had weak muscles and tones and stuff. So they said the best thing to do is to put a gastrostomy in."

Josh's mum talks about the difficulty of making decisions when a condition is unknown.

However, unlike Charlotte's parents who continue to question the appropriateness of a gastrostomy at a time when Charlotte's prognosis was unclear, Emily's parents and Josh's mum describe being able to draw on their child's subsequent diagnosis following the initiation of technological support, combined with the positive outcomes for their child, to manage the feelings of uncertainty they had experienced around the decision.

For Robert's mum, who has always considered the available prognosis when making decisions, the early uncertainty regarding Robert's prognosis meant that decisions made about particular treatments and interventions were made on the basis of a more optimistic prognosis than it was later realised by the consultants involved in Robert's care. This has led to decision regret for Robert's mum, who on reflection believes that some of those early decisions were not in Robert's best interests when taking into account the more realistic prognosis.

The cases presented here reveal the difficulties in being able to obtain a realistic prognosis with which to base decisions upon. They also draw attention to the unstable nature of life-limiting and life-threatening conditions, and as a result the changes in prognosis a family may experience during their child's life. Even for parents whose children have a well understood condition, prognostic uncertainty continues to feature in decision-making because of the individual progression in a child's condition. Parents can therefore find themselves in a state of continual uncertainty about the decisions they make for their child, both internally as they repeatedly ask themselves questions that cannot always be answered, and externally as they deliberate with others in an attempt to deal with the uncertainty they face.

"You just don't know.....Every time he was in hospital you think "is this it this time?" And he'd get better again and you think "right we'll battle on a bit longer.....you know is it going to make him any better? Will he be alright without ventilation? Will it help him? How long will it prolong his life? Will it prolong his life at all?" You know it's really difficult decisions to make aren't they? Coz you're thinking "what, what?" You just don't know what to do."

Peter's mum talks about the prognostic uncertainty regarding assisted ventilation.

8.2 The Individual Child

As well as considering a child's prognosis which brings into focus the progression in a child's illness or condition, parents stress the importance of making decisions

which are based on the individual child, and not just on what is known about their condition or the symptoms they experience. That decision factors are based around a child's quality of life, both in the present, the past, and in the future as parents and young people consider the potential impact of a proposed intervention highlights the fact that the decisions families are asked to make are not about the use of technological support per se, but are ultimately about their child.

The way in which the child features in decision-making is separated into three distinct yet overlapping categories – a child's physical and emotional resilience, in other words their ability to cope with the changes that a proposed intervention will bring; a child's views on the proposed intervention; and a child's developing capacity for decision-making.

8.2.1 Child's resilience

The intimate knowledge parents have about their child and about their child's physical and emotional resilience to cope with their illness was raised by many parents as they described their child's life, and feature in the on-going decisions families make for their child. It is not always an explicit decision factor, and can instead simply influence the way in which the potential benefits and drawbacks are understood by families. Either way, considering a child's individual resilience was identified as one of the strategies parents employ in order to assess how their child might respond to a proposed intervention.

"You see he has microcephalus, quadriplegia, epilepsy, spastic.....well Steven luckily, except fits, he's got away with the fits, they've been terrible fits some of them. But he's coped with them. And he has never had any big illness you see.....he's resilient you see. His kidney's good, his heart's marvellous."

Steven's dad identifies Steven's physical resilience to cope with his condition.

"By the age of about 14 months he got his first spinal jacket to help him sit straight.....but you know he was, he was quite a resilient little boy who, who still wanted to do everything and anything, and in amongst

that was quite, that did have some problems with, you know, chest infections, and very vulnerable on that one.”

Lee’s mum recalls Lee’s resilience to cope with his condition despite his vulnerabilities.

This was also described with regards to the ‘close calls’ children have experienced in the past, and the narratives of parents are interwoven with accounts of the near death events their child has survived. For parents, these events are symbolic of their child’s strength, and of their child’s continuous fight for life, despite the difficult circumstances under which they live.

“In the first three and a half years of her life, we went through eight times of thinking we were going to lose her.....Eight times we literally thought this is it, she’s on death’s door. But she amazed us every time and still does.”

Hollie’s parents talk about Hollie’s fight to live.

“The times he’s been put under is amazing. It says these boys you don’t really put them under anaesthetic but I couldn’t tell you how many times they’ve took him down.”

Alex’s mum identifies Alex’s resilience to cope with surgery, despite the risks involved.

Considering a child’s physical and emotional resilience to cope with the procedure itself, and to cope with the changes that the initiation of technological support will bring is especially important for parents of children with very severe cognitive or communication impairments. For some parents, this is seen as the only means of assessing how their child might respond, as their views are sometimes impossible to obtain.

“I was worried, what would Sebastian think of this strange plastic button on his tummy? And how would we explain that we would be feeding the food in. In the event, we explained it to him and he accepted it, and there was no problem. And some children try and pull it out, but he, there was no problem.”

Sebastian’s dad considered how Sebastian might respond to a gastrostomy.

Like Sebastian's dad, who described a process of 'viewing the world through Sebastian's eyes' in his attempts to understand how Sebastian might respond to a particular intervention, Hollie's parents relayed a similar practice, assessing their daughter's behaviour in order to understand how she coped with her feeding difficulties, even though Hollie was unable to offer any meaningful communication, verbally or otherwise.

Mum: "I mean she's emaciated, you know, she's like a stick insect."

Dad: "But if she wants to drink she will drink."

Mum: "She'll take it. That was the thing as well wasn't it, it's like it's a choice you know.....because I mean, how many times where I'd be there trying and getting her to do it. We can go for days where it's a real struggle and then all of a sudden she'll just start sucking.....it's like OK she wants it now, she's ready for it."

Hollie's parents identified drawing on Hollie's behaviour to understand her needs.

8.2.2 Child's views of intervention

The examples of Sebastian and Hollie draw attention to the role of a child's views in decision-making, even when their views are difficult to obtain, and parents are acutely aware that they are making a decision on behalf of their child that may cause distress and discomfort in the short-term despite the long-term benefits they identify. In fact, where possible, parents consider their child's views about a particular treatment or intervention when making a decision and this remained the case whether or not young people actively participated in the decision process.

The importance of including the child's views in decision-making is partly underpinned by the belief that where a child is happy to proceed, it is likely to lead to better outcomes.

"It was up to Peter whether he wanted to do it or not. If he'd have said "get lost I'm not interested", well what could you have done? You couldn't make him do it. You know, because there has to be a certain

level of patient co-operation with those, you know you can't just stick it on and say "right well that's it get on with it".....If they're fighting it, it's not gonna work."

Peter's mum describes the importance of including Peter's views in the decision.

Like parents, young people identified this feature in decision-making, and agreed that unless they are committed to a proposed intervention, especially one that requires them to make changes to their life it may result in poorer outcomes. A young person's views were identified as particularly relevant for decisions about assisted ventilation because of the cooperation required in using a ventilator, and the invasive nature of the equipment required. More generally though, parents and young people talked about the importance of having a positive attitude towards any proposed intervention, and referred to the impact it might have on their ability to recover well from surgery, or to cope with a new intervention that may have both negative and positive outcomes, or one that will require a lengthy hospital stay and recovery period, such as spinal rods.

"If they say do it and I don't want to then it's not really gonna work the same way as if you want to do it. If you want to be, you know, get better faster and lots of other things....if you don't want to do it then when you are trying to recover you're not really so determined to recover. You know, if you're forced into something you don't want to do, it's stupid."

Harry believes that recovery can be difficult when you are resistant.

However, distinguishing between a young person's views about an intervention and their immediate response to a recommendation was identified to be important, and in some cases a young person's initial resistance to a proposed intervention was due to a lack of understanding about the potential benefits, or fear regarding surgery or the equipment itself, as highlighted in Chapter Seven. For example, while Harry was optimistic about the potential benefits he would derive from having a tracheostomy, he nevertheless expressed fear during the decision process because

of the invasive nature of the procedure, which in turn led him to doubt whether or not to proceed.

8.2.3 *Child's developing capacity for decision-making*

Given the emphasis parents place on including their child's views in the decision process, it is not surprising that a strong theme in the data concerns the commitment of parents to involve their child in decision-making. The data revealed a continuum of participation for children and young people, with many children taking on an expanding role as their capacity for decision-making develops.

"You know when they're little, little, children, well you have to make decisions for them because they can't make their own decisions. But when they're older, teenagers and stuff, it's really difficult because they know what they want don't they? And then when Peter was eighteen he could sign his own consent forms and everything, because it was his decision."

Peter's mum reflects on Peter's expanding role in decision-making.

Parents are identified to have a key role in facilitating their child's participation, and the potential for young people to discuss their views is influenced by their parents' awareness of their ability and desire to do so, and their parents' views about how much say children should have in the decisions that affect them. In fact, parents are identified as the gatekeeper between their child and the decision process, allowing for different degrees of participation depending on the extent to which they believe their child will be able to process and weigh up the information required to make an informed decision, and whether or not their participation will cause distress or fear at a time when they are likely to be medically vulnerable.

Sam is a case in point here, for while he was able to voice concerns about the proposed technology, his actual role in the decision was constrained by his parents' views regarding his cognitive and emotional ability to make decisions, despite his developing capacity to do so. For Sam's parents then, their role was to inform Sam of their decision, providing him with information about a gastrostomy, and then to

reassure him that it was the right course of action in order to reduce the feelings of fear they identified him to have.

Matthew on the other hand has been involved in treatment decisions since around the age of twelve when he recalled being given a choice about whether or not to have spinal rods, deciding against it despite the procedure being strongly recommended by Matthew's consultant.

"They asked me if I wanted spinal rods in my back. And I chose to, I chose against it, I decided I didn't want that.....But it was me, the choice of me saying "I don't want it because of these reasons", and me standing by that decision that I'm not gonna have it."

Matthew describes having the final say in the decision about spinal rods.

Enabling young people to have a choice emerged as a key goal for parents, who identified the multi-dimensional role they assume in helping their child develop capacity for decision-making, and in facilitating their participation in the process itself. In particular, parents described their on-going efforts to ensure that their child is both informed, and supported in the choices they make.

"Where spinal surgery was concerned, Alex made that choice. I said I'm not making that choice. He had to go through it. And because they have to go through it, they have to make the decision. You just make sure they understand, if there's gonna be any side effects or anything. But whatever they choose, you just have to back them."

Alex's mum believes it is essential to make sure Alex is informed and supported.

Like parents, young people also make a distinction between the process of decision-making and the choices they make, and the young people in this study welcome the role parents continue to play in providing information, and feel enabled to make choices when they know that their parents will support the decisions they make.

"So they say [referring to parents] "even if we think you should do it and you say you don't want to, we aren't gonna say you have to do

it".....They told me that they'd never make me do it which actually means I've got a choice."

Harry recalls being told by his parents that he has a choice in decisions.

As well as ensuring that their child has access to the right level and type of information, parents describe assessing whether their child is able to process the information, and in some cases take steps to check that they understand the wider impact of initiating technological support. Other strategies include providing opportunities for their child to discuss the decision as a family, to meet other young people who use the technology, and to become increasingly involved in consultations so they are able to hear the opinions of the professionals involved.

"Don't give them too much information too soon, or too much. You know otherwise it scares them stupid and it's either that they think it's gonna be done tomorrow or they'll never ever want it done. And that's obviously you as a parent, you will gage it to the right level."

Lee's mum recalls making sure Lee had the right amount of information.

The role parents assume in helping their child make informed decisions and to increasingly expose them to the wider aspects of the decision process draws attention to the continual learning young people engage in. As a result, young people come to know the strategies they themselves can use to become informed, and to make sure they are enabled to make the right choices.

"I think because mum and dad have always asked lots of questions so I've sort of got it from them you need to do that.....So I've learnt it from watching them."

Harry identifies learning from his parents with regards to getting information.

In relinquishing decisional control, parents describe their responsibility to ensure their child is able to make the right choices. This, combined with the knowledge parents have from their own experience as decision-makers, motivates them to make sure their child is informed. This however can lead to some parents maintaining a degree of decisional responsibility because they themselves have

experienced the regret and other negative emotions when the outcomes of a decision have been poor. In providing support then, parents describe sharing decisional responsibility with their child in an attempt to reduce the feelings of uncertainty and regret that can sometimes occur.

In addition, some parents believe they have a role as ‘advisor’ that extends beyond that of ensuring their child is informed and supported, even when their child has full decision-making capacity and legal status as an adult patient. This is underpinned by the expertise and knowledge parents have of their child, and the advice they believe they can therefore offer on this basis.

“I talk to him and I say right, this is how it is. I’ve thought about it. And Alex will sort of in some respects disagree because he always thinks for himself, but as he tells me, he trusts me. And because he trusts me and he knows that I wouldn’t put him anywhere or do anything that would put him at risk then I always say to him, look Alex if I’m not happy with something, which I’m allowed to, as a mum, to express that.....And sometimes that’s what he needs. And then when he realises, he thinks about it and he goes “hang on a minute you’re right”.”

Alex’s mum describes her role as advisor to Alex.

There was also evidence in the data that one particular element of the decision process, that of prognosis, is sometimes withheld from young people, even those with fully developed capacity for decision-making. Both Harry’s dad and Lee’s mum revealed having separate conversations with their child’s consultants to talk specifically about the decisions in terms of prognosis, and to discuss the implications of technological support in relation to its ability to prolong life.

“I used to do a lot of talking in front of Lee, but sometimes I would make an appointment by telephone or I’d physically go and see them and say, I can’t talk about this, you know the prognosis, in front of Lee. I can’t be as honest.....I just did not want it rammed down Lee’s throat because he

was living life to the full, and he was loving everything that he did.”

Lee’s mum felt that discussing Lee’s prognosis was not something Lee could cope with.

This is not surprising given the findings presented in Chapter Seven that young people focus on minimising the impact of their condition on their lives when considering the benefits and drawbacks of technological support. Moreover, the life-shortening nature of their condition was identified by some young people as a difficult subject to think about, something that was also recognised by parents.

“To me it was trachee or die. Well it was really. There’s no ‘ifs and buts’ about it.....I mean I didn’t think much about why I should do it. I just did it coz I thought well “I don’t like being ill”.”

Alex admits not thinking about the potential impact of ventilation over the long-term.

“You know what’s gonna happen don’t you. I think they just don’t want you to talk about it, they maybe just want to ignore it really, and live for the moment, which is what they do. You know, maybe they just don’t want to discuss it with other people.”

Peter’s mum believed that Peter did not want to think too much about his prognosis.

This can have an impact on a young person’s ability to adequately weigh up the benefits and drawbacks of a proposed intervention over the long-term. Therefore, for some parents assuming control of this aspect of the decision is part of the overall process of assessing their child’s capacity for decision-making.

There are other circumstances under which children can move back and forward along the continuum of participation and this can be related to the complexity of a decision, a child’s diminishing capacity due to the progression of their condition, or a child’s physical and emotional resilience at the time of the decision being made. Consequently, the level of control parents assume in decision-making will also change as their child’s capacity to make decisions moves back and forth.

Among the families in the study there were several examples of decisions being made during an acute illness, which limited a young person’s capacity to participate

in the decision process. The decision for Alex to have a tracheostomy is a good example of this scenario and the life-threatening nature of his condition at the time a decision was reached about a tracheostomy meant that Alex felt in no state, emotionally or cognitively to make an informed decision, and was happy in this case for his mum to do so on his behalf. However mum in this case still made an effort to involve Alex, and due to her intimate knowledge of her son she was able to draw meaning from the verbal utterances and facial expressions he could make at the time a decision had to be made.

“His eyes told me everything. And I said “do you understand what they’re saying?” And he said “yeah”. And I said “they’re gonna just put a trachee in and then you know, you can come forward”. And he’s “yeah” he was like “yeah”, I could communicate with him enough but they [referring to professionals involved] didn’t understand what he was saying.”

Alex’s mum describes her role as translator at a time Alex was unable to communicate.

This case provides an insight into the ability of young people who experience an acute illness to weigh up the decision they are asked to make, and Alex admits that his understanding of a tracheostomy at the time of making a decision was incorrect as he believed that he would no longer require the ventilator after the mask was replaced with a tracheostomy.

“At the time I was like, it was very hard to picture it. I didn’t really know what would happen. So when I had the trachee in I thought I was just breathing oxygen because there was no mask. I didn’t know you could put a ventilator into your neck. I didn’t know you could use this [indicates to ventilator] with a trachee.”

Alex misunderstood the information he was provided with to make a decision.

For Alex, an agreement had already been made between Alex and his mum about who would take control of decision-making at those times when his capacity might be constrained by an illness, and this was also the case for Matthew and Kate, who

discussed with their parents the possible decisions that might be made during this time.

“Alex always says that if he can’t make a choice and he’s under anaesthetic or whatever, then I make the choice. He knows that whatever I’m gonna do will be in his best interest.”

Alex’s mum recalls discussing her involvement at times when Alex lacks capacity.

Overall then children and young people gradually assume control over the decisions about their condition as their capacity develops. Nonetheless, in all but one case, parents and young people continue to share the process of weighing up the benefits and drawbacks of a proposed intervention, and to some degree the choice being made. The following passages illustrate the importance to young people that their parents are involved in the decision process, and the key role parents assume in helping them to understand the potential impact of the treatments and interventions being recommended.

“My mum and dad all go together to all the things, we all go together.....We’ve always made choices together. We found out all the information, and mum and dad have always told me things as far as, well told me things that I’d understand.....coz different ages you understand different things.....and then in the end I decide in the end what the best thing is.”

Harry talks about a shared process of decision-making.

The exception to this is Richard, who was the only young person in the study living independently. Richard’s experience of decision-making is not one that is shared with his family, although it has been in the past and Richard will sometimes still discuss decisions with his parents in the process of choosing whether or not to proceed with a particular intervention.

“I mean definitely my parents always have, they’ve always allowed me to voice my opinion on decisions but I’ve always been quite strong

mindful in terms of wanting procedures to benefit me. So really my parents have never had to say "oh well yes", or talk about it and stuff. They've just signed the document for me."

Richard describes taking full decisional control quite early on.

8.3 Parents as Decision Makers

Within the discussion of young people's developing capacity, it is clear that parents play a central role in facilitating their child's participation in decisions that affect them. However, like children, parents embark on their own journey as decision makers, and this in some ways resembles the journey children make in that parents also develop capacity for decision-making over time, and the parents in this study provide remarkably similar accounts of moving from being dependent on the healthcare professionals involved in their child's life to assuming control of the decisions that need to be made for their child.

"The journey we've had with Hollie's situation has been probably like all the others, phenomenally emotional, an emotional rollercoaster. Up and down and round and round. And the first three years were probably the hardest and there after it's got a little bit easier."

Hollie's dad describes their journey as parents of a seriously ill child.

Looking back, many parents described themselves as "completely naive" at the time their child is diagnosed with a life-limiting condition. By this, they refer to their ignorance in terms of disability, their lack of knowledge regarding their child's condition, their limited awareness of what support they might need, and their lack of understanding about how the healthcare system works. Parents also describe themselves as being "overwhelmed" by emotions having recently been informed that their child has a life-limiting condition, and for new parents this can come at a time when they are already exhausted from the changes associated with becoming a new parent.

Consequently, parents recall having to rely heavily on the medical professionals they have access to at this time to advise them on the best course of action to take when making decisions for their child. They describe having little choice but to place their trust in the medical establishment, which some parents had only previously encountered as a child, and then to give birth.

“That was the start of all these, all this sort of millions and millions of professionals that you become overpowered by.....so if you have a problem, right that’s it, shove a load of professionals your way, and you know, and then you don’t know who you’re talking to or what’s happening.”

Lee’s mum was overwhelmed by the professionals involved and how to deal with them.

From this state of vulnerability, parents undergo a transformation as they gradually assume responsibility for their child’s care and treatment. This is partly achieved through the efforts parents make to become informed, but is also shaped by their growing knowledge of their child, and the expertise they acquire as primary carers. It is also influenced by their encounters with the healthcare system, in other words the process factors that will be discussed in Chapter Nine, and many parents report developing an awareness that the recommendations they are asked to consider may not always be in their child’s best interests, and that the system as a whole perhaps does not work as well as it could.

“If you don’t know they provide you with what is convenient for them to give you, but if you do know that what you’re provided with is probably what they’ve got, not what is best for the patient, then you can do something about it.”

Sebastian’s dad describes his growing awareness of the flaws in the healthcare system.

In assuming responsibility, both as parents of a child with a life-limiting condition, as carers of a child with sometimes very complex healthcare needs, and as decision-makers for the on-going management of their child’s condition, there are three key themes identified from the accounts of parents as they describe their role in

decision-making:- becoming informed; gaining control; and making the right choices.

8.3.1 *Becoming Informed*

In an effort to reduce the overwhelming emotions they experience as parents of a newly diagnosed child, many parents describe their on-going efforts to become 'informed', and this is a central stage of the journey parents make. In fact, for some parents becoming informed is the single most important aspect that enables them to become a good decision-maker and carer for their child. Learning about their child's condition, about the possible treatments, and about where help and support might be available, enables parents to ease their feelings of uncertainty and fear, and equip them with the necessary knowledge for their new roles as 'carer' and 'decision-maker' for their ill child.

"For us, what was scary was not knowing. Knowing removes the scariness. What was scary for us was being in the dark, and not knowing that the light at the end of the tunnel wasn't actually the other end but a train coming towards us. You need, we needed information, and that was the major problem."

Sebastian's dad describes the role of information in reducing uncertainty.

There are three main ways in which parents become informed. Firstly, they arm themselves with as much knowledge as they can acquire about their child's condition. For some parents, this can happen quite early on in their child's life. However, for others there is more of a gradual realisation that the responsibility they as parents shoulder for making the right decisions for their child is theirs alone, and this can drive parents to become experts in their child's condition. Becoming informed is not an easy process though, and many parents describe it as frustrating and time-consuming.

"I mean knowledge is a jigsaw, and you have bits of information and slotting it in so you get the overall picture. And if there isn't anything to hang it on then very often there's a gap....So it was hard work and very,

very frustrating.....because we realised that there's a lot more information that's not accessible."

Sebastian describes the difficult process of acquiring the information they needed.

However, for some parents their past experiences in both their personal and working lives can mean that they know where to look for information and who to ask. In fact, it was evident in the data that there is a lot of variation in the types of information parents draw on in the process of becoming informed, with some parents accessing scientific journals regarding the latest advancements in their child's condition, others seeking out the best possible medical expertise in the country to obtain information from, and some making do with the information available from professionals, condition-specific charities, the internet, and other families.

"Perhaps my life experiences, previous life experiences helped me. If I'd have been a 22 year old coming across this for the first time I think I would have been completely at sea. But because I was an older dad, and had a certain background."

Sebastian's dad describes the importance of his former experiences.

The second way in which parents become informed relates to their dual role as parent and carer, and parents report gaining knowledge of their child as a person, and developing expertise regarding the impact of their child's condition on their child's health and quality of life. As the main carers of their child, parents often become tuned into the minutia of their child's health state, and are able to detect the slightest change in the medical indicators that lead to a recommendation for treatment or medical intervention. These clues can be so subtle, and some parents gradually become aware that only they are sometimes able to detect them.

"We were able to pick up on posture, body language, facial expression, eyebrows, mouth, eyes, breathing rate.....I could tell if the muscles were in spasm. I could feel the bubbling in his chest, see if he needed aspirating. I could feel the rhythm of his breathing, whether it was

relaxed or whether it was a bit hasty. I could tell whether he was cold, hot, sweaty, clammy."

Sebastian's dad describes his in-depth knowledge of Sebastian, as his primary carer.

Again though, this journey is not easy and parents recall times during which their growing confidence and expertise can falter, with some parents drawing attention to the physical and emotional exhaustion associated with providing around the clock care. For other parents, these feelings of self-doubt can occur when their child requires a new medication or treatment, or the initiation of technological support.

"Well I couldn't do it, so this community nurse said to me "I think you can do it". I said "oh no, I'm not. I can't, you know I'll kill him because I'll put the wrong, I'll mix up the wrong amount. It'll be too dangerous." So she'd come in the morning and then at night. She said "I think you can do this. I'll see you through it.""

Lee's mum recalls times where she felt out of control.

While discussing decisions about technological support, parents identified the difficulties inherent in the dual role as parent and carer, and the emotions they have to overcome in order to carry out what can be distressing or painful tasks on their child. As a result, parents can require on-going support from medical professionals as their ability to cope waxes and wanes, and their child's care changes over time.

"They showed us how to sort of, it was scary at first when you see your child with it, you know, having a hole in there [refers to tracheostomy]. And then you know, you get used to it and they show you how to change it. And I remember the first time they took it out and they wanted me to do it. I couldn't do it. You know, it was just, you think "I'm gonna hurt my child"."

Josh's mum recalls her fears as she took over the care of Josh's tracheostomy.

Finally, parents talk about how crucial it is to be informed with regards to the on-going decisions they make, and of making sure that they are able to access the

information and advice they need to make the right choices for their child. Many parents go to great lengths to ensure they have the information they need for decision-making. Nevertheless, parents are yet again dependent on the professionals in their lives if they are to have access to good quality information that enables them to make informed decisions. This will be discussed in Chapter Nine, and both the involvement of professionals and access to information for decision-making were identified as key process factors in the model.

“You have to have all the information to make the right choices, especially if it’s a child, for your child. So you have to fight. Don’t just sit back and take their word for it. You have to fight.”

Casie’s mum on the importance of being informed when making decisions.

8.3.2 Taking control

Over a period of several years parents describe being transformed into ‘experts’ regarding their child, their child’s condition, and the necessary care they require. This transformation also shapes their role in decision-making, because being armed with this knowledge and expertise enables them to gradually assume control over decisions that will affect their child’s life. Ultimately, by taking control parents describe gaining a sense of control over the situation they find themselves in as parents of a child with a life-limiting condition.

“I mean every day you come across things, and you have to make decisions. But you just get on with them and you overcome them. When you’ve got a child who’s ill or who has a disability, you know it’s part of life. And you just make decisions at the time that you think are best.”

Josh’s mum on becoming the decision-maker for her disabled son.

“Rather than being a victim and feeling that you’re at the mercy of circumstances, you can take control and become a manager. And although there are difficulties, because you feel that you’re managing the situation, the whole situation becomes different.”

Sebastian’s dad describes the process of gaining control.

How long this process takes will vary, and Robert's mum recalls taking control during the middle years of Robert's life as her knowledge and expertise grew, and she came to realise that as Robert's parent and primary carer, she was the best placed person to make decisions on his behalf. She also believes that being a confident and assertive person made it easier to assume control in decision-making, and to advocate on Robert's behalf.

"At some point, once you realise that you have a better idea of what's wrong with your child, and how serious it is, you can say "well actually, we respect your opinion but on this occasion....".....we grew to trust ourselves to make the right decisions for Robert."

Robert's mum recalls the point at which she gained control of decision-making.

However, this transformation is not always easy for parents, some of whom describe having to become assertive and confident, traits that they are not naturally comfortable with, but that they recognise to be important in the process of assuming control over the care and treatment their child receives. In addition, parents describe the on-going fight to ensure their child receives the right care, and this can be exhausting and difficult to keep up at times when parents are feeling emotionally vulnerable or tired.

"You just have to fight the whole way through. You learn how to become assertive and to say not, you know, and not feel bad for saying "no I'm not happy about this".....you've got to have that self-belief to say "actually this is my child, this is your patient yes, but this is my child. They are the centre of my universe and I know you're caring for them but I love them".....but it's hard to have that fight when you're so tired."

Lewis's mum describes the on-going fight to get Lewis the care he needed.

By gaining control, the dynamics between parents and professionals can change, and many parents recall becoming more vocal during consultations and hospital stays to ensure their child receives the care they need. Parents also realise over time, as they themselves acquire competence and expertise in managing their

child's condition, that not all professionals have the same level of expertise, partly because of their unfamiliarity with children of this complexity. Consequently, parents are more likely to challenge the care their child receives, and ensure that the professionals and carers involved in their child's life have the necessary knowledge to carry out their child's care efficiently and safely.

"It's sort of knowing her. You know. One of the times she were at the hospital she were having her fits and she were chesty.....And we just walked through, and we just went "coming through". I says to him "she needs to cough, that's what's up with her". Coz her colour were really grey. So I were there with the catheter, he pressed her throat, she coughed and she were fine. Pink in two minutes."

Emily's dad describes challenging the care Emily was receiving in hospital.

Charlotte's parents point out that with expertise and information come the confidence and knowledge to challenge the opinions and recommendations made by healthcare professionals, and to question whether the care and treatment pathways that are recommended are in Charlotte's best interests. Parents' increasing knowledge and control therefore alters the dynamics for decision-making, and parents sometimes begin to question the authority and expertise of the professionals involved in their child's life.

"We had to really fight against it. Saying, look this is not what we want, and stand our ground. And sometimes you think, you know, "I'm only his mum, should I be listening to you [consultant]" and "they know better". But sometimes I think "they don't". You know your own child."

Matthew's mum identifies her increasing expertise and control over decision-making

"Obviously there's a few I trust. But professionals, I put that in a bracket because at the end of the day, they're no more professional than us as a family. You know I don't go in thinking, I've cleaned my boots, I'm sat next to a professional here. Well he might have digits and whatever against his name but he will still never understand this condition. I've

lived with it for 22 years and I still find it difficult.”

Alex’s mum compares the expertise she has to the professionals involved in Alex’s life.

In fact, one of the reasons parents find it so important to become an expert and take control of decision-making for their child is the gradual awareness that some medical professionals hold information back, and that in some cases a decision has been reached among the professionals involved about a gastrostomy or ventilation before the point at which it is ever suggested to a family. Therefore becoming informed is for some families a way to check if all the options that are available are being presented to them.

“So they don’t start off with “this is the issue and these are the options and we recommend this one”. They start off with “this is the issue and this is the way we deal with it”So just make sure that you’ve got all the facts, not just the ones that are presented to you.”

Harry’s dad believes not all the available options are always presented.

8.3.3 Making the right choices

In assuming control for the decisions that must be made for their child, parents consistently refer to their child’s best interests when reflecting back on the decisions they have made, and are committed to ensuring that the choices they make are ones that will enhance their child’s quality of life. In fact, parents’ accounts of the decisions they make are repeatedly discussed with reference to their child’s quality of life and their commitment to ensuring that the choices they make are in their child’s best interests.

“From our point of view, our responsibility was really to make, was the duty of care, that we had to make sure that we looked after him as best we could, as any parent would. And if that involved going to a lot of trouble then so be it.”

Sebastian’s dad emphasises their duty of care to Sebastian as his parents.

Through the process of becoming informed and gaining control, parents are better able to understand the quality of life their child can achieve, which Sebastian’s dad

admits can be hard to do unless you have an intimate knowledge about your child and their condition. Therefore, having this knowledge can help parents to determine whether a treatment or intervention being recommended is in their child's best interests.

"Our focus was, obviously, what was in Sebastian's best interest. And it's sometimes very difficult as an adult to put yourself in the position of a child. But there's a child there who needs you."

Sebastian's dad admits it can be difficult to assess what is in a child's best interests.

Deciding on this basis also offers parents reassurance that when the decisions being made are in their child's best interests, the risks are worth taking, even when they are significant.

"There are risks with everything. When they said to us it would take an operation, like you know, there's no guarantee whether he'll come back or not, it's just one of those things you have to take but like I said, you have to take the risk don't you, in the end because you've just got to think what's best for your child."

Josh's mum talks about having to accept the risks involved.

"If I made the decision then it was on my back.....but I can sleep at night if it's in Alex's interest that I do something, then I do it."

Alex's mum is committed to making the right choices for Alex.

Parents are acutely aware of their responsibility as decision makers for their child, and continually ask themselves if the decisions they make are in their child's best interests. Some parents describe the potential conflict they experience in considering an intervention that if initiated will have mixed consequences for their role as carer. A gastrostomy is a good example here because once initiated the hours spent feeding their child each day may come to an end, yet the new regime of care associated with a gastrostomy will replace it, accompanied by the emotions parents can experience around the loss of oral feeding.

“That would make our lives easier, or mine particularly because I was the one who was spending all the time feeding her and then mopping up after her.....but it wouldn’t make Hollie’s life any easier and we thought of the tube in all the time, that she may never feed again orally.....so we weighed all of this up because we wanted to make sure that what we were doing had Hollie’s best interests at heart, not ours. And if it meant that she was going to die then so be it you know.”

Hollie’s mum talks about making sure decisions are in Hollie’s best interests.

In fact, the data revealed a strong commitment among parents to make decisions on the basis of their child’s quality of life, and although they are often aware of the potential consequences for themselves as parents and carers, these consequences are viewed by all the parents interviewed as something that they will have to cope with when the time comes.

“You don’t want to be getting right invasive with your own kid unless you’ve got to. But it were just something else we had to cope with.”

Emily’s dad on taking over the care of Emily’s increasingly complex needs.

Ultimately then, parents wish to make decisions that will benefit their child. Yet for decisions about technological support their ability to do so is compromised by not only the uncertainty regarding the outcomes, but also by the uncertainty regarding their child’s condition in terms of how it will affect those outcomes, and with regards to the progression of the condition itself. The commitment parents take on to ensure they make the right choices for their child can therefore be overwhelming, and Alex’s mum admits it is sometimes easier to make a decision in which there is no perceived choice, because there is more certainty that the choice taken is the only viable option, and consequently the right decision.

“We just didn’t have a choice, and that’s not a bad thing at that point because it was basically, you say to yourself “right”. You say to yourself “there isn’t a choice here. If he doesn’t have the trachee put in, then we haven’t got Alex anymore”. So there’s no in-betweens. There’s no, let’s

read some information and see what's what."

Alex's mum admits that sometimes it is easier to make a decision where there is no perceived choice.

Hollie's dad also refers to the emotional burden of making such complex decisions, and admits that it is sometimes difficult to challenge the opinions and recommendations being made, especially when you are being told that what is being proposed will make your child feel better. Therefore, even when there is no perceived pressure in the decision process, parents can still sometimes find it easier to agree because the consequences of choosing the option not to proceed requires a great deal of effort on the part of parents to make sure that by going against a recommendation, they are doing what is in their child's best interests.

"This is the problem when you're going through this, there's so many emotions going through your mind because there's, you know, whatever's going wrong is going wrong with your kid. To have somebody tell you "we'll do this and it'll make it better" is you know, of course you want to go for it."

Hollie's dad describes the challenges of making decisions for your own child.

Therefore, although parents describe a journey of becoming informed and gaining control, which in turn enables them to feel confident of their ability to make the right choices, there are still particular decisions, or particular circumstances under which parents question their ability to make informed decisions for their child, and in this scenario parents can require more support and input from trusted professionals.

"And we're exhausted at that point, absolutely at the end of our tether, you know, and then making these decisions when you're in that state. In this totally, mentally, physically exhausted state, and that's not taken into consideration all the time."

Lewis's mum draws attention to the emotions and fatigue that can affect their decision-making capacity.

Finally, one of the consequences of gaining control and becoming informed is that parents sometimes begin to reflect back on earlier decisions that have been made, and for some parents this can come with the realisation that certain choices may not have been in their child's best interests. Reflecting back on decisions of this nature can be emotionally distressing, because in some cases parents recall the strength of their own desire to keep their child alive at the time of making a decision, which forces them to consider whether or not this may have come at the expense of their child's quality of life. Because many of these early decisions are sometimes taken on the advice of the professionals involved, parents begin to question the agendas and biases within the recommendations that have been made, illustrating the interactional nature of decision features and process factors in the model.

8.4 Beliefs and Values

Inside the narratives of parents as they described the process of gaining control as decision-makers for their child, are the evolving beliefs and values that both shape the journey they take, and are in turn shaped by their experiences of parenting a child with a life-limiting condition. By reflecting back some parents were able to identify the role of these beliefs and values in decision-making, but at the time had not realised that their decisions were partly underpinned by them. In other families, parents report developing an awareness of their beliefs and values over time, as they assume responsibility for making on-going decisions for their child. Young people too were able to recognise the role of their beliefs and values in decision-making, and like parents identified the evolving nature of their beliefs and opinions, which for young people are also shaped by their experience of living with a condition that impedes their quality of life.

In particular, the life-limiting nature of a child's condition, and for some parents the 'close calls' their child may survive prior to making decisions, can come with a growing awareness of their child's fragility for life. Young people identified the impact of these life-changing events as well, and meeting head-on the life-

threatening nature of their condition can force young people and parents to confront the views they may have about life and death. Beliefs about the role of fate and faith in determining the outcomes of decision-making were also detected in the data and for some families can influence the way in which decisions are made. The influence of other beliefs were identified too, shaped by the on-going encounters parents and young people have with the healthcare system, which bring into focus the perceived biases and barriers in the choices presented to them.

8.4.1 Guiding principles for treatment

Beliefs and values can act as guiding principles for parents to draw upon when considering treatment and other medical interventions, and some parents shared with the researcher their views on life and death, and on the purpose of medical intervention when discussing the role of technological support for their child.

“The biggest thing is our attitude, and our change as we realized there is nothing we can do about it other than be there for her, allow her to be as comfortable as possible and that she doesn’t belong to us. You know, you don’t own people. You care for them and when she’s ready she’ll do her own thing.”

Hollie’s dad describes his changing attitude towards Hollie’s life.

The study revealed a continuum along which parents are positioned with regards to their approach to treatment, and the above passage illustrates the fluid and evolving nature of these positions as a child’s illness progresses. At one end of the continuum, there are parents who will do whatever it takes to keep their child alive.

“So you’ve got to kind of say “well yes”, because you just clutch at anything don’t you, to keep them alive. You know, you just clutch at anything they offer you that might just prolong their life that bit longer.”

Peter’s mum on her treatment philosophy to do whatever it takes.

At the opposite end, there are parents who believe in allowing a condition for which there is no known cure to progress at its natural rate without unnecessary intervention.

"If it meant she was going to die then so be it you know.....it would be sort of, under her terms, as opposed to you know us prolonging it by feeding her artificially or you know doing anything like that. It would be a more natural progression of things."

Hollie's mum talks about her treatment philosophy for Hollie.

The position parents assume can shift over time, influenced by their experience of parenting an ill child and the knowledge they develop regarding their child's condition and prognosis. As well as changing as parents become more informed, there can be certain times or events during which these positions are called into question. Some parents may re-consider their approach to treatment following the outcomes of a particular intervention, which can alter the expectations parents have for their child's quality of life. Others adopt a temporary position along the continuum during a holiday without their child, in case there is a sudden or acute deterioration in their child's condition during their time away. Experiencing the death of their own parent or another close relative can also challenge parents' beliefs and values about life and death, which again can influence the approach to treatment they adopt for their child.

Some parents are forced to make decisions about life-sustaining technologies very early on in their child's life, before they have had time to get to know their child or to understand the severity and nature of their child's condition. In these cases their existing values about a life worth living are all they have to refer to, and these values may be abstract and vague, and not previously used to inform decision-making. Some parents admit that regardless of the values they might have held in the past, in reality they are sometimes little more than useless when one of the decision options will mean it is *their* child who will die.

Lewis's parents found themselves in this situation, and describe how difficult it was to make a decision for Lewis, partly because they were unable at this stage to assess the sort of life he would be able to achieve.

"We had to make a decision about the ventilator didn't we?.....And it was a case of well you know, we take him off it and if he doesn't breathe on his own or carry on, you know, there's a decision to be made type of thing. Whether we kind of put him back on it or we just let nature take its course.....Well I think we were both a bit, I think I was more inclined to let nature take its course. I think you wanted [refers to Lewis's dad] to put him back on and, it was all a bit difficult. I think it was the first time we had to make that decision."

Lewis's mum described having to consider their approach to treatment very early in Lewis's life.

Indeed, views about quality of life influence treatment philosophies, and some parents describe in detail their commitment to ensure their child has the best quality of life they can achieve. For some parents of children with muscular degenerative conditions, this can take the form of keeping their child as physically mobile for as long as possible. In these cases, children may undergo a number of medically invasive procedures to achieve this goal. For others, the philosophy to do all it takes means becoming informed about their child's condition, and keeping up to date with the latest scientific research and clinical trials.

"It's always been like there are things that we can work on to make it as good as possible. So it was always done in the light of "look, it's not as if there aren't any options anywhere for anything and that whatever options are there we'll get them for him". So although clearly very bad news, it was always done in a way "so we've got to get together and make the best of it."

Harry's dad describes their approach to managing Harry's condition.

Sometimes though, it is simply about ensuring a child is given the opportunity to achieve their goals and dreams, and Lee's mum described her commitment to provide Lee with as many opportunities as possible in what they both knew would be a short life.

"Like he should try everything and anything, and my god, did I try and live up to that, you know. So he did try everything, and anything he wanted to try and do, he did."

Lee's mum describes focusing on life and living.

Young people also talked about their treatment philosophy, and their views on life and death. However, their views were more consistent than those of parents because their desire to stay alive and focus on their ability to participate in life influenced their attitude towards interventions that were ultimately life-prolonging. Rather than paying attention to prognosis in terms of the number of years left, interventions like a gastrostomy and ventilation were instead seen as life-sustaining, and in some cases life-enhancing.

In fact, choosing life was a dominant theme, and young people in the study admitted finding it difficult to think about the life-limiting nature of their condition when making decisions of this nature. Alex describes this approach, choosing life in every case no matter what the procedure is, and instead finding ways in which to maintain his quality of life, despite the inevitable progression in his condition and the impact of the additional equipment required to support him.

"I think the only thing I would say is just do whatever keeps you alive. As simple as it sounds, that's sort of logic that isn't it? If it keeps you alive I don't care what it is, because in the end it doesn't matter what you've got. You can still do things so you've just got to find a different way of doing it.....You've got to keep going so you can see your family. Life is it, that's it, it's life. That's the only information I had – life. My own information, you know, but if it keeps you alive then you've got to do it."

It's life or, well I don't really think about anything else."

Alex admits finding it difficult to think about his prognosis when making decisions.

Ultimately, the proposed technologies sustain or prolong life even when decisions are primarily about preventing malnutrition or tackling the effects of sleep apnea. Even when decisions are not viewed by families as 'end of life' decisions, the life-limited nature of a child's condition and the uncertainty regarding their prognosis draw attention to further deterioration in a child's condition, to the life a child has left, and the impact the proposed technology may have on that. Therefore the values and beliefs held by families about the purpose of life and living are central to the decisions that are made for children with life-limiting conditions.

8.4.2 Beliefs about the influence of fate, faith and luck

This category is not about the role of fate, faith and luck in decision-making. It instead refers to the way in which an individual's beliefs about the potential influence of faith, fate and luck over the decision process can affect how parents and young people weigh up the use of technological support, and how they assess important because they are sometimes drawn upon by families as a way to reduce the uncertainty experienced by families during the decision process. The influence of beliefs that individuals hold about fate, luck and faith in decision-making was not explicitly identified by all families, although in the decision narratives they were revealed among some of the families who did not identify this as a factor.

Some parents discussed the role of religion in decision-making, and talked about drawing on their faith to re-assure them they are making the right choices for their child, and that the outcomes will be favourable. For Harry's parents, their faith helped them to cope with the uncertainty surrounding their decision for Harry to have a tracheostomy, and the risk of something going wrong. Their particular decision was made against the recommendation of several consultants, and their faith was an important factor in helping Harry and his parents sustain their belief in what they felt was the right choice.

“As a Christian you believe there’s somebody behind it but I mean you can’t write it. You couldn’t write the script.....If we’d made the wrong decision then, it would have been a totally different situation.”

Harry’s dad talks about the role of faith in decision-making.

Sebastian’s dad also identified the role of his faith in the decision regarding Sebastian’s gastrostomy. Like Harry’s family, it was a way of coping with the uncertainty involved in the decision, and of having the faith that all would be well despite the risks involved of agreeing to a gastrostomy.

“If it’s going to improve the quality of life for the child then, and there’s obviously the risks involved in the gastrostomy, but assuming that your child comes through it, and you have to have faith in the skills of the surgeon and the operating team.”

Sebastian’s dad talks about putting his faith in the surgeon due to the risks involved.

Other parents talked about the role of ‘luck’ in determining the outcomes, and in the same way that some parents draw upon their ‘faith’ to manage uncertainty, others make use of their child’s ‘close calls’ to cope with the uncertainty in the decisions they face, describing how lucky they have been that their child has survived thus far.

“We did just sort of, we’ll take it as it goes really, you know. Even with adults and children, it’s just life. So you just sort of have to think well what’s best you know and hope that everything comes back alright sort of thing, and that they’re well.....I mean so far, life’s been good for us really, I can’t complain.”

Josh’s mum believes they have been lucky so far.

However, not all families feel lucky, and this belief too influenced decision-making. For example, Hollie’s mum recalls being influenced by her belief concerning their ‘bad luck’ as a family when interpreting information regarding the potential outcomes of a gastrostomy for Hollie.

“He spelled it out very clearly from what I remember of it. Even down to staying there’s 10% of kids who have this who don’t get any benefit. Because I remember thinking welcome to the 10% club. That’s what went through my head when he said that. That will be us, that’s where we’d be, in the 10% you know.”

Hollie’s mum recalls believing that, given their previous luck, a gastrostomy would not work.

Where some parents put their faith in god, and some parents hope for the best, others take a more fateful approach to decision-making, believing that the outcomes are already determined. Like luck, this approach is shaped by the journey their child has already travelled, and is again utilised in part to manage the uncertainty regarding their child’s condition and the decisions they are asked to make.

“I think in myself I knew that, I knew this wasn’t his time to go, and I didn’t know why. But I think there was a reason in my eyes, because I always think things happen for a reason.....Nobody knows why they’ve happened. They’ve just happened and nobody’s got an answer for it. So now we just go with the, with the flow, whatever’s gonna happen will happen at the end of the day.”

Alex’s mum describes her views on fate and life.

8.4.3 Beliefs about the health care system

Beliefs about the influence of fate, luck and faith are sometimes used by families in considering the potential outcomes of a proposed technology. In contrast, the views and beliefs parents and young people express about the health care system tend to influence their opinions of the professionals they encounter, the way in which the recommendations made for their children are interpreted, and the way in which the information they receive for decision-making is reviewed. One of the strongest themes here relates to a general feeling among both parents and young people that the choices they are presented with are already constrained, and that they are not always made aware of what is available.

“Lots of people just accept what is said to them and that’s it. I mean like I say, some things aren’t for everybody, and we’re all different, but we know that, we’re not stupid. It doesn’t work for some people, it does for others, but you’ve got to try it before you know if it’s gonna work. And if you don’t get that chance of trying it, how do you know it’s not going to work?”

Harry’s mum believes that not all the available options are always shared with families.

“They provide you with what is convenient for them to give you.....So we realised this early on, that we weren’t being told things. So we researched everything, and then said, oh by the way....”

Sebastian’s dad realized that they were not always offered what was available.

“They actually do know more than they’re telling you.....and you can’t always take what they believe as gospel because it’s not always true. Not that it’s not always true, but that it’s not exactly as they say it coz it’s a bit like, it’s kind of spin really isn’t it?”

Harry shares his concern that consultants don’t always tell you everything.

Many of these views are influenced by prior encounters families have experienced as their child continually dips in and out of the system. However, some of the beliefs and views are drawn from a family’s existing knowledge about the NHS, and from the on-going media and political debates concerned with limited resources, funding cuts, and poor standards of care.

“Back in the day before the NHS, if you went to see a doctor you had to pay. And you know, the doctor, a physician, a doctor, it was an art. Which they, now they, it’s all machines. They take, you know, they don’t look at you now. They would have a look in your eyes, they’d have a look at your tongue. But doctors don’t do that now.”

Hollie’s dad believes that the NHS has changed.

“We hate any sort of invasive surgery in the UK.....It’s like the hip thing [refers to hip replacement for the elderly]. Let them be falling over

before we do the hip.....They're obviously nervous about operations and you know, being sued as well, coz if it goes wrong, well there was another option."

Harry's dad shares his views about the approach to surgery adopted in the NHS.

Some parents talked about the attitude within the system towards disabled children and the treatment of children with very complex health care needs. These beliefs affect the way parents interact with the system, and can force them to take a different approach when they feel something needs to be done for their child, especially when it is felt that their needs are given a lower priority when compared to other patient groups.

"But for children like this, if you don't shout up for them they'd be left. They'd be at the back of the queue every time, money-wise and everything."

Steven's mum shares her views about the treatment of disabled children.

"My theory is if you're a patient and wait your turn and you keep quiet you wait forever. And if you shout and make a noise you get help, eventually. Become a nuisance.....Threaten national papers, make a fuss."

Sam's dad believes that you have to push to get what is needed for your child.

"What part of him having a voice don't they understand? That he has you know, it's this whole, coz you have a physical disability or something. It's like you're deaf and you're dumb. It's crazy. Like they just see him there and think he's dead already.....And it's awful, to see that disability thing still hasn't moved on."

Alex's mum shares her views about the approach to treating disabled people.

The views parents shared about the NHS also influence how they view the professionals working for the system, and there was a general impression across the families in this study that you were fortunate if you were able to surround your

child by the right people, who it was felt went above and beyond what is expected of them in order to make sure a child receives the treatment and care they require.

“Anybody who says the NHS works hasn’t experienced it properly....I reckon 80% of the staff who work in the NHS aren’t that interested....They turn up, do their job, go home, forget about it. The other 20% actually care. But you’ve got to find them.”

Casie’s mum shares her opinion about the NHS workforce.

“By the time they’ve [nurses on the ward] done 12 hour shifts. If it’s full on, by time you’ve got to 10 hours they’re absolutely zonked. It’s too long.....Three days....Three 12 hour shifts. 36 hours that’s it. Get it all done in three days.....but then if you got a good nurse, where you were confident with, and you thought, bloody hell she’s good.”

Sam’s dad discusses staff shifts and the perceived impact on quality of care.

“Well we had this wonderful, wonderful woman who, yet again, went 110, 115 per cent of the way....I think I was quite fortunate from that point of view, not like other people had in the area, or in other areas.”

Lee’s mum felt lucky that Lee had dedicated professionals involved in his care.

“I think some of them, some of the top surgeons like to perform in front of a crowd. He had students there and people from around the world had come to specially see him doing his work and, yeah, he seemed quite a showmanship, which some of them do have.”

Richard has the impression that surgeons are sometimes driven by other agendas.

It is clear from the narratives of parents and young people that the influence of their direct experiences with the healthcare system, and the role of media and societal views of the NHS and the treatment of disabled children are intertwined and therefore difficult to disentangle. However, whether or not they are influenced by families’ direct experiences, it is essential to view the beliefs families have as a decision feature because they can influence the way in which a recommendation for technological support is perceived, with some

parents believing that they are made on the basis of the resources and priorities within the system rather than what is in the best interests of the child.

When families believe this might be the case, there is potential for conflict regarding the decisions that are made, and for Molly's mum her impression of the NHS led her to believe that Molly was discharged from hospital before she was ready to go home following her gastrostomy and fundoplication surgery. Molly died the following day after being re-admitted to hospital, and the feelings of anger and blame continue to cause distress several years later as Molly's mum continues to try and make sense of what happened at the end of her daughter's life.

"They sent her home the day after, which we thought were really early because she hadn't even really had anything down it [refers to gastrostomy]....I know half that ward were closed at the time so they were needing beds and stuff....but then that night she were just, it were just all weeping, but it took till the next day to get her back in coz they had no beds. And then that's when it all went wrong.....she had a massive fit and the intensive care doctor refused to treat her coz he said she didn't have any quality of life....but I just think that's wrong because if you've got a handicapped child you think that if they make a mistake, or something goes wrong, that they should try and make sure that child's alright, no matter what."

Molly's mum describes the events leading up to Molly's death.

8.5 Gut Instincts

This section is about the role and influence of gut instincts in decision-making, which is identified as partly an emotional response to an initial recommendation for an invasive or cumbersome medical intervention, and partly an instinctive response to a recommendation that may also be informed by the growing knowledge assumed by parents and young people in their developing role as decision-makers.

Referred to by families as a 'gut feeling', a 'gut reaction', an 'instinct', or 'intuition', it is for some parents and young people a central feature in the decisions they make, and one that is hard to reason with even when they are aware of the emotional basis of the reaction they experience.

"You know, they said "well look he's not gonna make it. You know, there's a possibility of just turning the machine off." And I'm thinking you know, it didn't really, it didn't feel right. I know it didn't..... I'm thinking "he's got quite a lot of life still to go yet.....I don't know but I'm going by my gut instinct here." If something doesn't feel right then I don't go in that direction."

Alex's mum recalls her gut reaction to agree to a tracheostomy.

Parents reflect on the strength of this initial response, which some refer to as a gut feeling or reaction, and others as an 'instinct' that is hard to overcome, even when the recommendation or information goes against what their instinct is telling them. For Molly's mum, the strength of her gut reaction led her to cancel a gastrostomy three times, as she continued to insist that it was not in Molly's best interests despite the persistence of Molly's consultant and those around her, including her long-term partner who had been a father to Molly since she was an infant.

"I just didn't want it. I don't know, I just didn't want it doing. I knew that I didn't want it doing. And I thought, I don't know, I thought "there's something not right about stopping some-one from being sick if they need to be sick." I don't know. I just, I don't know. I mean the thought of her never being sick again were lovely but I just didn't want it doing.....I was adamant I wasn't having it done. I were really adamant."

Molly's mum describes her gut reaction to the recommendation for a fundoplication.

Like Molly's mum, Robert's mum refers to her instincts when making decisions for Robert, again drawing significance from them. She goes on to recommend that all parents should trust in their gut feelings regarding a decision, even though in Robert's case not all of the decisions made on this basis have led to positive

outcomes. Alex's mum too identifies the important role of gut feelings, and when asked what she would say to other parents faced with a similar decision, she recommends that parents should "go by your gut instinct. You'll know if the decision's right".

However, the role of instinct in decision-making is complex. Some parents distinguished between the emotional and rational basis of these gut feelings, referring to the initial fear and anticipated loss they can experience when faced with decisions that involve risk or that may cause their child distress. This is compared to the influence of their growing knowledge and expertise in forming an initial response to a recommended treatment or intervention, which they believe is more rational and informed than the early gut feelings they experience as parents of a recently diagnosed child, when they know little about their child or their child's condition.

"It's about the whole, everything that's gone before, you know, and your gut instinct as well. You know coz we just knew that it was [pause] we had to let him go. And we'd fought so hard for him, on so many occasions."

Lewis's mum describes drawing on both her gut feeling and her knowledge of her son.

Young people also identified the emotional and rational basis of decision-making, and Harry explained that sometimes a gut feeling is nothing more than an emotional response to a recommendation, and therefore can at times be "irrational". Again, young people refer to the information and knowledge that is required in order to challenge these 'gut reactions' and make a more informed decision.

"There's always an initial sort of yes or no, initially, a yes or no. Because I know with the tracheostomy at first I thought "I don't want to do that. That's a stupid idea, why would that happen".....So at first you get your own feeling straight away, an initial reaction, and then they give you the

proper decision-making tools to make your choice properly.”

Harry talks about gut reactions in decision-making.

Nevertheless it can be difficult to tease apart these feelings, which in some cases can influence how parents and young people interpret information about the risks involved in a proposed intervention, and how they assess the potential outcomes. However, families emphasised their commitment to make objective and rational decisions for their child, words they themselves use to describe decision-making, despite the emotions involved. Indeed the journey parents embark on as decision makers for their child, and the growing ability and capacity of young people to make decisions enable them to gradually acknowledge the role of gut feelings in decision-making, and to distinguish between instincts that are based on the expertise and knowledge they assume over time, and the emotional gut response to an intervention being proposed.

8.6 Chapter Summary

The themes presented in this chapter describe key features about families of children and young people with life-limiting conditions that can have an impact on the way in which families approach decision-making, and the way in which they interpret the recommendation and information they are given in order to weigh up the appropriateness of the proposed intervention. Although this chapter presents specific features about the patient population, the findings reveal that the features are not static, and instead evolve over time as parents and young people make on-going decisions about their treatment and care. Sometimes their influence in decisions is identified by families, however in some cases the role these factors play in decision-making is implicit and difficult to detect, and families are not always aware of how these factors influence the process of decision-making.

Chapter Nine will now examine the category of process factors, in other words the factors regarding the healthcare system with which families must engage with when their child's quality of life is impeded by a symptom or associated problem regarding their condition for which medical intervention is required.

- CHAPTER 9 -

PROCESS FACTORS

This is the final chapter of findings and provides a detailed account of the category of process factors included in the new model of treatment decision-making developed from the research. The process factors discussed in this chapter relate specifically to the healthcare system within which treatment decision-making occurs, and therefore includes the role of professionals, and the use of information for decision-making.

The chapter is divided into four parts. The first is concerned with the decision background, which contains important clues and indicators about the relationship families have with professionals at the time a decision is being made and the time families may have already spent in considering the use of technological support. The second section concerns the role of professionals in decision-making, and the third considers how information is accessed and interpreted by families as they weigh up the decision being made. The chapter finishes by considering how the on-going interactions families have with the healthcare system influence their evolving role as decision-makers and on their views about the NHS more broadly.

9.1 Background to Decision

Chapter Eight provides an account of the journey parents embark on as decision-makers for their child, and for some parents the background to decisions about technological support is an important and influential part of that journey. Indeed the events and encounters families experience in the period leading up to a recommendation were viewed as an important part of the overall decision process, and this information is crucial if we are to understand a family's views, knowledge, and position at the time a decision is being made. It also draws attention to the range and type of professionals who may have been involved up to this point, and the level of agreement that has been reached between these professionals and the

family about both the symptoms a child is experiencing, and the solutions available to manage them.

“Sometimes, they just forget the background that you’ve come from. Do you know what I mean? And we would only go to hospital when it was desperate because we would try to avoid going to hospital coz it was much better to care for him at home.....So at that point you get to hospital, you’re at crisis point, and I don’t think people always take that into consideration, that the whole background of how you came to that point. So when you are involved in all these decisions, and also people don’t always take into account your specific knowledge of your child. So in some cases, they’re sort of going through the motions of telling you x, y, z, and you know more than they do.”

Lewis’s mum describes how essential it is that professionals take the time to understand the background to decisions.

The background offers vital details about the decision context, and can provide useful information to professionals about why families have particular views and opinions regarding the decisions they are asked to make. As the case of Sam shows, the background to the decision about a gastrostomy was particularly important because it revealed the pressure Sam’s parents felt placed under to agree to a gastrostomy quickly as a result of their strong desire for Sam to have spinal rods, and to have the procedure during the window of time that it would benefit Sam the most.

The background to the decision Casie’s parents made also contains important clues, offering an explanation as to why they felt so angry at the time a recommendation for a gastrostomy finally came, and when compared to the decision made for Sam, portrays the very different circumstances under which recommendations for technological support are made.

“Every time I spoke to the neurologist I said she was choking on her milk, she had swallowing problems.....And I mentioned it every time didn’t I? I

made a point of mentioning it because I was at my wits end.....And it was only when we were in hospital, however many months later, while I was getting fed up with everything that was going wrong and people not listening. And I actually asked to see the notes, and she'd made no mention of swallowing difficulties. I was absolutely stunned."

Casie's mum describes some of the events leading up to the decision.

Having knowledge of the background to the decision is therefore essential if we are to understand how the related experiences encountered so far influence the way in which families will react to the recommendation for their child to receive support from a life-sustaining technology, and how they will respond to the professional making the recommendation. It can also provide an indication of the information and support families might need at this point and the amount of weighing up families have already done. As Chapter Seven highlights, some families may have already reached a decision about the initiation of technological support at this time, whereas others have no prior indication that a recommendation for technological support will be made.

This is not surprising considering that for some children, parents must make decisions about artificial nutrition or assisted ventilation very early on in their child's life, or for older children following an acute admission to hospital which then reveals the need for technological support. However, in many cases children have swallowing or respiratory problems over a sustained period of time before they are considered for artificial nutrition or assisted ventilation, due to the progressive nature of their life-limiting condition. During these months and in some cases years there is a difference in the type and number of professionals involved in a family's life, and in the relationship a family has with them.

For some families, their child's difficulties are not acknowledged by professionals for some time despite parents informing them of their concerns about their child's health and associated quality of life. For children with feeding or swallowing difficulties, there is often an on-going 'battle' to ensure their child receives adequate nutrition. For children with respiratory problems, parents can be in and

out of hospital with their child as they suffer from recurrent chest infections. This, combined with a child's deteriorating health, and the lack of acknowledgement and support families report receiving from the only people who can help is not only distressing, but has the potential to affect their developing relationship with the health care professionals involved in their child's life.

For other families though, their child's on-going swallowing or respiratory problems are not only acknowledged, but monitored on a regular basis as the family, together with the professionals involved, attempt to understand what is wrong and find appropriate solutions. Sometimes, a gastrostomy or ventilation may be discussed over a period of years. For other families, it may not be mentioned as a potential solution until a child reaches the point at which it becomes very clear they are unable to receive adequate nutrition orally, or to breathe without assistance. Nevertheless, when a child's problems are acknowledged, parents describe feeling supported and reassured that they are not alone in managing their child's symptoms, and that the professionals involved are working with them to resolve their child's difficulties.

There was some evidence in the data that acknowledgement is more likely for children and young people with conditions where it is expected there will be feeding or respiratory problems, especially when compared to children with undiagnosed or very complex conditions. For these families, a child's swallowing and respiratory function is sometimes monitored for years, and the decision was described by some families as a planned one, gradually introduced to families at different time points before being initiated at the appropriate time. In fact some young people recall always knowing they would need ventilation and therefore accepting it as inevitable, and do not always identify having made a decision about ventilation per se, but rather about the timing, type, and location of initiation.

However, this distinction does not always apply, and there were examples in the data of children with well understood conditions whose symptoms were not acknowledged by the professionals involved. There were also families in the sample whose child's condition was unknown and their prognosis difficult to determine, yet

whose feeding or breathing difficulties were acknowledged and monitored. When a child's symptoms are not acknowledged, some families will seek the opinion of other professionals and make attempts to initiate the decision process, taking decisive action and demanding that their child be considered for technological support. However, for other families a child's difficulties continue to be unacknowledged until a life-threatening event brings to light the extent of a child's swallowing or respiratory problems.

By examining the background to decisions about gastrostomy and ventilation, common pathways through which children come to be considered for gastrostomy insertion and assisted ventilation were identified. These are summarised in Table 4 on the following page (p. 255), which portrays the very different journeys families make as their child experiences the symptoms leading to a recommendation for technological support. Each journey gives an indication of the difficulties families might have experienced at the point a recommendation comes, and the potential conflict they may have encountered with other professionals along the way. It can also offer an understanding as to why parents might present as angry and emotional, or resistant at the time a recommendation is made.

9.2 Time Period

The very different backgrounds identified in the study portray the variation in the time period that is available to families as they consider whether technological support will be of some benefit. At one extreme, the process is gradual and subtle, taking place over many years so that when a recommendation comes, the decision has already been reached, and some parents and young people in this scenario were not able to recall actually making a decision. At the other extreme, the decision process is so short that families do not always have time to consider the range of benefits and drawbacks of the proposed technology, and instead the decision is made on the basis that their child needs some form of medical intervention because of the severity of their symptoms, and the sometimes life-threatening situation they can find themselves in.

Table 4 – The Background to Decisions: Case Studies

Background Characteristics	Case Study
<p>Uncertain and Unacknowledged</p> <ul style="list-style-type: none"> • Acute episode likely • Involvement of known and unknown professionals • Access to information variable • Lack of regular monitoring 	<p><i>Casie’s decision was made after a long period of time during which Casie’s parents encountered various professionals, many of whom would not acknowledge the progressive nature of Casie’s condition or her feeding difficulties. This led to a period of hospitalization after an acute illness. It was at this time a gastrostomy was recommended by unknown professionals with limited knowledge about Casie’s underlying condition.</i></p>
<p>Uncertain but Acknowledged</p> <ul style="list-style-type: none"> • Acute episode possible • Involvement of known professionals • Good access to information • Regular monitoring 	<p><i>Lewis was surrounded by a team of trusted professionals who communicated honestly with the family about Lewis’s prognosis. There was a lot of uncertainty about his undiagnosed condition and it was difficult to predict what quality of life Lewis could have. Nevertheless, Lewis’s parents felt well informed and supported by the professionals involved, who monitored Lewis on a regular basis.</i></p>
<p>Expected but Unacknowledged</p> <ul style="list-style-type: none"> • Acute episode possible • Involvement of known and unknown professionals • Access to information variable • Lack of regular monitoring 	<p><i>Matthew has a well understood condition and his respiratory difficulties were being monitored on a regular basis by a known professional. However the wider impact of his respiratory problems was not acknowledged by this professional and it took the involvement of other professionals, also known to the family to initiate a decision. Matthew therefore had access to information from different sources.</i></p>
<p>Expected and Acknowledged</p> <ul style="list-style-type: none"> • Acute episode unlikely • Involvement of known professionals • Good access to information • Regular monitoring 	<p><i>Richard’s decisions were made over a long period of time during which his swallowing and respiratory problems were expected to worsen, and were therefore being monitored regularly by known professionals. Because of this, Richard had access to a range of information for decision-making, and was able to make decisions that were not based on a life-threatening situation or acute illness.</i></p>
<p>The Crisis</p> <ul style="list-style-type: none"> • Short time period • Acute episode • Involvement of unknown professionals • Limited access to information 	<p><i>Alex’s decision exemplifies the situation families can find themselves in when there is a sudden and life-threatening situation requiring hospital admission. Families in this case can be surrounded by unknown professionals, who may have little knowledge about a child’s condition, and therefore little advice and information to offer a family for decision-making.</i></p>

“It was more or less, you know, it had to be like, a snap decision.....or he probably won’t make it through the night if you don’t.....you know and we had to decide really like right now, not next week or the week after. Just like sometime within the next hour.....so it was a bit of a forced decision. Well you know we didn’t really have time to think about it. It hadn’t been discussed or debated.”

Peter’s mum on the urgency of the decision regarding ventilation.

The decision process may or may not begin at the point of recommendation, and can span a period of minutes as was the case for Alex who was admitted to hospital with a pneumothorax and needed to be ventilated, or last weeks and even months as families take their time to think about whether technological support will benefit their child, as was the case for Lee who took his time to weigh up the pros and cons of a gastrostomy. For some families, the time period can change during the decision process depending on a child’s condition or the circumstances under which the decision is made.

“There were talking about having a gastrostomy and saying “oh you know it’ll take about four months to have done, and then it became four weeks, and then it actually happened in four days.”

Casie’s mum describes the changing time period over which the decision was made.

From analysing the data, there a distinction between the overall decision process, which can last many years as a child’s symptoms worsen and parents continually assess their child’s needs, and the actual process of deciding, the latter of which is defined as the time during which families weigh up the appropriateness of technological support, and acquire the information they need to make an informed decision. For some families, the duration for these different aspects of the decision process is the same, and this was certainly the case for families who had not expected a recommendation for technological support to be made.

“That [gastrostomy recommendation] came in sudden because we were expecting him to have his back operation....and then “oh we’re gonna

put him in for this instead”.....And of course you just think “I hope they haven’t gone and messed about and left it too late”.”

The overall decision process and the process of deciding were the same for Sam.

For some families, the time in between making a decision and the actual initiation of technological support is also included as part of the decision process, because it provides families with another chance to re-consider the choice they have made.

“We initially left the hospital saying yes but we went away, we sort of, because we weren’t that convinced about it. But we said “we’ll say yes and go and really think about it”. Coz you know it’s easier to turn round then and say no, than it is to say no now and then say yes later because it will have to start all over again.”

Hollie’s mum used the time after saying yes to think about the choice they had made.

For children who are already in hospital, this period of time can be quite short so as to avoid a lengthy stay in hospital, and this was one of the reasons the time period for Casie’s decision shortened so dramatically. Josh’s mum also identified the difference of making a decision during a hospital stay, and welcomed the fact that once she had decided to proceed with a tracheostomy and gastrostomy, steps were taken to organise the surgery very quickly.

“It was kind of one of those decisions, because he was already there, kind of a matter of days. I wasn’t left to ponder over it for months and months, which was good really. I mean once the decision was made they could get him into surgery because he was there already.”

Josh’s mum welcomed the short period of time between making a decision and surgery.

In contrast, many of the children and young people who are elected for technological support as part of a routine consultation have a longer wait between the decision and the initiation of technological support, and some parents recall their child being placed on a waiting list. Therefore, the period of time between deciding and initiation is variable and can span several months. This can be distressing for parents who, having made a decision must now wait while their child

continues to experience what can be distressing symptoms. Given the uncertainty in these decisions, parents can sometimes spend this time questioning whether or not they are making the right choice for their child.

However, regardless of the circumstances, families welcome having enough time to make an informed decision, and young people in particular value being given time to think about whether or not they wish to proceed, to be able to discuss the options with their parents and in some cases other professionals, and to access the information they require to fully understand how technological support will impact on their life.

“You can’t just decide on the spot because some of these things are big decisions.”

Richard believes having enough time to decide is essential.

“I mean it’s not as if like, she needs it now, this second. You’ve got time to think about it which was helpful because you’ve got to chew it over.”

Emily’s mum welcomed having some time to think about a gastrostomy.

9.3 Professional Involvement

Both the background to a decision, and the period of time families have to make a decision can affect the number and range of professionals families will have access to during both the overall decision process, and the actual process of deciding. Some children have a multi-disciplinary team of professionals around them, who may have supported the family for years and who will sometimes discuss with the family the possibility of a gastrostomy over a sustained period of time. This can include nurses, speech therapists, dieticians, paediatricians, specialist consultants, physiotherapists, and other medical and non-medical professionals.

“There was a specialist nursery that was held in the hospital grounds, dealing with children with quite complex needs really. It was wonderful, the staff, do you know what I mean? And the physio used to come and the OT, and the speech therapist, and everybody who seemed to be

anybody....the consultant used to come down and grovel around on her knees, you know how amazing was that?"

Lee's mum described being surrounded by a good team from early on.

Other families may have one or two professionals involved with their child on an on-going basis, but who nevertheless believe they are well supported. For others, the decision is made with a team of professionals who work closely together on a hospital ward or in a specialist clinic.

"You see he basically spent about 18 months of his life in PICU [Paediatric Intensive Care Unit] completely. So all the doctors that were there at the time and surgeons, we were all sort of discussing it with them.....I remember the consultants being involved, the nurses being involved, and us being involved. It wasn't just the case of the consultants. It was the whole team. The staff were wonderful."

Josh's mum identifies the team of professionals who supported them.

These examples contrast greatly to the family whose child is surrounded by unknown professionals at the time of making a decision, particularly when they have limited experience of working with children who have complex and life-limiting conditions, which can be the case for children who experience an unexpected acute episode requiring hospitalization, and for children who encounter new professionals as part of their routine care who are unfamiliar to a family.

"I mean he can eat fine, and you know all these other people who're involved, they never though he needed a gastrostomy. And they've been involved for a long time, a long time. She was new. She just didn't know him you see. I mean some people have got a gastrostomy, you know they need a gastrostomy. But that's a different thing you see."

Steven's dad believes that professionals who don't know Steven do not always see the bigger picture.

Families with limited access to professionals who understand their child and their child's condition, and families who make decisions at the time of their child's

diagnosis may have few resources for decision-making, and are sometimes entirely dependent on the professionals involved at the time to make the best decision for their child.

“Your only information source is the consultant who has his own agenda, especially like, because I was staying in hospital with Casie I had no other source.”

Casie’s mum identifies the limited resources for decision-making from professionals.

In cases where unknown professionals are at the centre of the decision process, families will sometimes involve members of their own care team, especially when they have developed close and trusting relationships, and there are many examples in the data of parents and young people actively contacting their consultant for advice about whether or not they should agree to technological support. There were also occasions on which these individuals were asked to advocate for families, and in extreme cases to argue the family’s case when a family disagreed with the recommendation being made. For others, a children’s hospice or charity can become involved, and there were again examples in the data of individuals from such organisations taking on the role of advisor and in some cases advocate.

Families therefore differ in the decision-making resources available to them, and there is potentially a range of professionals who may be involved at different stages in the process who are not necessarily limited to the medical and allied health professions. Considering the journey parents and young people embark on as decision-makers, and the evolving relationships they form with professionals, this aspect of the decision process varies hugely, and is complicated further by the perceived agenda, biases and traits parents perceive some professionals to have, and many families describe a process by which they learn which professionals to trust and who to take advice from.

“They’ve got their own ideas in their heads. They are slightly biased on their own side aren’t they?.....When you listen you can’t always take what they believe as gospel because it’s not always true. Not that it’s

not true, but it's not exactly as they say coz it's a bit like, it's kind of spin really isn't it? They spin it in their own direction because they think that's right. And it doesn't always make it right. But they think it is."

Harry talks about professional biases.

Within the accounts of families as they discuss decision-making are rich descriptions of the many professionals families will encounter over the years, and both parents and young people draw reference to their character and style, and their approach to interacting with children and parents. Nearly all of these descriptions relate to the paediatricians and other consultants families meet, who are identified as the key resource for decision-making, and one of the main sources of information. Although parents highlight the key role of community nurses and allied healthcare professionals in providing invaluable support and advice, and in enabling families to access other professionals and services, it is the individual consultants who parents mainly identify with regards to the recommendations for technological support, and the process of making a decision.

Given the mixed experiences families have with the professionals they come across, parents and young people emphasise the importance of building a network of trusted professionals around them. However, they admit that this can be difficult to achieve, because sometimes finding the 'good ones' can take time. In particular, professionals who take the time to listen, who take the time to get to know a child and focus on their quality of life as well as their health, and who take the time to get to know the wider family and understand how everything fits together, are the professionals who are valued the most, and ultimately who families trust to give informed advice and recommendations.

"The true professionals are the ones who are friendly, become a member of the family without being over-bearing or intrusive. They're there in the background. They offer support and advice. They are very skilled. They're often intuitive."

Sebastian identifies the skills he identifies with a good professional.

From analysing the narratives of parents and young people, three key characteristics were identified that exemplify the professionals families value the most, and who make a difference to the overall care a child receives and the decisions that are made – seeing the child; acknowledging parents; and being accessible and flexible.

9.3.1 Seeing the Child

In describing the style and personalities of the professionals that families interact with, it is clear that where professionals engage with children and young people in a meaningful way, parents and young people are more likely to trust them, and to listen to their advice and opinions.

“Doctor C were lovely. I’ve always liked him from day one. He’s got a right soft accent and I could listen to his voice all day. But he, he always used to like ruffle Molly’s hair and say “alright”, and always talk to her. And if he were asking me anything he would say “is that alright Molly?” You know. He’s always been like that. He’s always consulted her.”
Molly’s mum recalls the efforts of the consultant to involve Molly.

Peter’s mum identified the informal approach taken by Peter’s consultant, and valued greatly the fact that the consultant spoke mainly to Peter, and treated him like an individual.

“He used to laugh at Peter because he used to sit with one leg crossed up on top of the other you know, and he used to say “how do you manage that?” I said “is he supposed to do that?” He says “if he’s comfortable let him sit how he wants”.”

Peter’s mum commented on the informal approach the consultant took with Peter.

Considering that decisions are underpinned by a child’s quality of life, it is not surprising that where professionals take the time to get to know a child and to listen to their views, this is highly valued by parents and young people because it signifies a commitment on the part of the professional to develop an understanding about a

child's quality of life. By doing so, parents believe that professionals are more likely to make recommendations on this basis.

However, both parents and young people draw attention to the fact that not all professionals will take the time to get to know the child, or to seek their views, and Richard has found from his experience, that sometimes you have to speak out for yourself in order to be heard.

"I think you need to be quite confident in coming forward and express your views to the medical professionals because sometimes they might not ask you."

Richard feels that sometimes he has to be quite assertive to be heard.

Richard also draws attention to the way in which professionals interact with him, and believes that it is important to make children and young people feel at ease so they feel confident enough to voice their opinions.

"I think they should build up a rapport and try and ask a few personal questions, and try and be quite chatty.....Build up a relationship and make sure you feel at ease and then go into the actual consultation. And then once you're in the consultation you might feel easier to express what views you have."

Richard identifies a key role for professionals in establishing relationships.

Some families discussed in particular those professionals with specific experience in working with children who have a life-limiting condition, who they believe not only have greater expertise and knowledge, but also a greater understanding of what life can be like for children. Drawing on this observation, Richard believes that his consultant is more committed to getting to know him and listen to his views than other professionals he has come across with less experience.

"Consultants that know you very well and have maybe experienced a lot of people with your condition. They know that your view is quite, that this is going to change your life quite a bit, so it's up to you really."

Richard identifies the consultant who understands the bigger picture as the one who will also take the time to listen to your views.

9.3.2 Acknowledging Parents

As well as acknowledging the individual child, professionals who acknowledge and respect the growing expertise of parents, and the responsibility parents shoulder to make the right choices for their child, are valued greatly by parents, who in return respect the professional's own expertise and knowledge, and take the time to establish a relationship with those professionals who approach families in this way.

"The professionals that talked with you rather than to you were the ones you could trust."

Robert's mum identifies a shared approach as crucial in building relationships.

"He'd proper like talk to me, like he were my mate. And there's not many consultants you can do that with, that would be happy for you just to ring them at work, randomly out of the blue. But he were fine with it and I was saying "I'm sorry to ring you", and he'd say "oh it's no problem"."

Molly's mum identified the informal relationship she had with Molly's paediatrician.

However, when parents encounter professionals who fail to do this, they can be left with feelings of frustration and anger, and parents draw attention to the potential conflict in the developing relationship with that person, and with other professionals who they subsequently meet as a result of those early unpleasant encounters.

Mum: "The first paediatrician she saw when she were four months old, she told me to go out and get a job."

Dad: "Neurotic mother."

Mum: "Because when you have, when it's your first child, sometimes you see things that aren't always there. So she told me to go out and get a job."

Dad: "She told us that there were nothing with our Emily's head."

The knowledge Emily's mum had of her daughter was dismissed by a paediatrician.

Building trust therefore takes time, and for families who have experienced difficulties in having their opinions and growing expertise acknowledged, professionals will sometimes have to work hard to develop a trusting partnership with children and parents. Nevertheless, many parents reported that some professionals intuitively know how to interact with parents, and relationships that begin from a point of mutual respect are those valued the most by parents, who will utilise this person above other professionals involved in their child's life where possible.

"It so happened that we went to the hospital and saw a chest specialist, who actually was excellent as a consultant. He had an excellent style, and he made it clear, this is a co-operative effort. "I have some expert knowledge but you have expert knowledge" And he said "you can provide me with as much information, which is new to me, as I can the other way round". So he made it clear that he wasn't an expert, he was an expert at not only giving us information but listening. So he was incredibly useful. He did all sorts of, shall we say he was flexible with his interpretation of the rules in order to get Sebastian what he needed."

Sebastian's dad describes what he sees as a good professional.

Finally, in acknowledging the expertise and knowledge of parents, professionals must also be aware of the journey parents take as decision-makers, and therefore recognise that the ability of parents to take control comes with time, and can fluctuate depending on the circumstances of a family at the time a decision needs to be made. This can be seen in Lewis's case, who discussed their contrasting opinions about one of the consultants involved in Lewis's life who at the time Lewis's mum found overbearing and opinionated. However, over time as she gained control as a parent and decision-maker for Lewis, she came to respect his honesty, and the information and advice he provided.

Dad: "Whatever his opinion was, and whatever he thought were gonna happen or not happen, he just told you straight. Which at the time was exactly what I wanted...."

Mum: "...I was too fragile, you know what you're like when you've had a baby anyway....he was very abrupt. He was very much of that he would explain things to us and then he would sort of make doubly sure that you understood.....and in that very early point where you are so fragile anyway, it was just too much for me to cope with."

Lewis's parents describe their different needs at the time Lewis was diagnosed.

9.3.3 Being accessible and flexible

Many parents describe the professionals they can rely on as those who go above and beyond what is expected of them in order to meet the needs of their child, and parents compared them to other less competent and less willing professionals they had encountered in their efforts to get across to the researcher how grateful they were for what was sometimes a single professional that the family could rely on, and that would "go the extra distance" for children in their time of need.

"She was his paediatrician, consultant you see. She delivered him and she's known him all his life and she's stuck to him.....I mean last time he were in hospital she said "you get in that car and go home." He was asleep, he were unconscious. She said "go home". I said "right".....She's great, I mean she'd just come back from her holiday, she weren't really on duty but she come in and she says "I'll see to him now"."

Steven's dad identifies Steven's paediatrician as the key professional in his life.

"He's just such a nice man, would do everything that you, he'd always, always, always went extra, and he was fantastic and he would even tell me when he was gonna have time off, you know "right I'm going away on holiday so I just needed to tell you that so and so is going to be in my place if you need anything"."

Lee's consultant wanted to make sure Lee could access help when he was on holiday.

In “going the extra mile” for families, which was a dominant theme in parents’ accounts of the professionals they value the most, parents are describing professionals who are both accessible and flexible, which for many families is seen in stark contrast to the wider healthcare system in which they work. Many parents expressed a need to know that they can contact some-one who knows their child in the case of an emergency, and this is not surprising considering the progressive and fluctuating nature of childhood life-limiting conditions.

Having the mobile phone number of their paediatrician or another consultant is highlighted by many parents as important, and offers reassurance that if they need assistance or advice, they have some-one they trust a phone call away. Being trusted to use the personal number of their child’s consultant with discretion also signifies to parents that their own expertise is acknowledged in the relationship with this professional, and some parents talked about making sure only to use the number they are given in the case of an emergency in order to respect the partnership that is established.

“At the end of the meeting he pushed his card across and he said, and he turned it over and on the back was written his home telephone number. And he said “you’ve a difficult time, any problems ring me at home.” Now that obviously is the sort of person you want. Not some-one who thinks, I am, you know, mini god.”

Having a personal number of the consultant was important to Sebastian’s dad.

Giving parents a personal number also implies to parents that this professional is flexible in their approach to care, and this is essential for families who recall occasions during which their child has been unwell at times when accessing help is difficult, for example on bank holidays and during the night. Offering support in a flexible way that considers the individual needs of each family is therefore hugely beneficial, and enables families to dip in and out of the system when they need support, and to tailor the support to meet their individual family’s needs.

“The consultant supported us straight away. He just said, basically “what can I do?” And we just said “we’d like to come every now and again just to talk it through.” He knows there’s nothing he can do anyway but he’s happy for us to go there. He listens to her chest and gives her a little check out. And it’s just nice to know that he’s there, and we can talk it through with him. And that if we need that help we can give him a shout.”

Hollie’s dad emphasises the importance of having access to some-one.

These professionals are also seen as a family’s anchor in the wider healthcare system, acting as gatekeepers and advocates as they opens doors to further support to ensure a child receives appropriate care. So as well as being flexible and accessible in the advice and care they provide directly, these professionals are sometimes involved in helping families access other interventions, support, and equipment a child might need from other services, and in some cases bending the rules to achieve this.

“In the end it was the chest physician. He looked at Sebastian and said “are there any other problems connected with breathing?” And we mentioned that he wasn’t feeding well and he said “well this is dead easy”. He says “just get a dietician”. So he rang up and a dietician arrived within half an hour.”

Sebastian’s dad identifies the role of professionals in helping to navigate the system.

Families vary in the level of support they have, and families who described being well supported will in most cases have an inner circle of trusted professionals who they know well, and who they trust to do everything in their power to put their child’s best interests first. For some families, this may only include one or two professionals, even when they have access to a wider range of professionals who are known to them. This illustrates the fact that merely having access to professionals is not enough for parents to feel supported, nor to have appropriate resources available to them for decision-making.

“So if we’ve got any queries or questions or anything we get in touch with G [community paediatrician] and she directs us in the right, either in the right direction or twists the right arms. But it’s really important to find as many people as you can who will do that for you because it’s the only way you get what you want and it’s the only way you make, you stand the chance of making the right decisions, by getting all the information that you want.”

Casie’s mum identifies the role professionals have in enabling parents to make informed choices.

9.4 Information for Decision-Making

There is a distinction between the information that is available to families for decision-making, and the information that families actually need to make an informed decision, and this relates to the interactional nature of the model in that the information families need for a specific decision is influenced by the journey parents make in assuming control of the decisions regarding their child’s condition, which in turn is shaped by their on-going interactions with professionals, who are identified as the primary source of information for decision-making.

Where families have access to trusted professionals who are flexible and accessible, the information available to them for decision-making is greater than for families who are surrounded by unknown professionals. In addition, families who are proactive in seeking out their own resources for information are less likely to depend on professionals to provide all the information they need.

Therefore, the information and support families have access to at the time of making a decision about technological support is influenced by both the care team surrounding the family over the period of time a decision is made, the individual professionals directly involved in the decision itself, and families’ own resources for information. The time with which families have to decide also has an impact on the amount and type of information they are able to access. Finally, the decision background is relevant here, because where some families may know very little

about the technology being recommended, others may have already sourced a range of information at the time a recommendation is made.

As parents and young people discussed the process of weighing up the appropriateness of technological support, they identified different types of information, which they accessed from various sources including medical professionals, other families, charities, and the internet. Table 5 on the following page (p. 271) provides a summary of the main types of information identified from the data, and the main sources families utilised to access the different types of information they use in making decisions.

9.4.1 *The recommendation*

For most families in the study, the ultimate recommendation for a child or young person to have a gastrostomy or ventilation comes from either the child's own paediatrician, or another consultant that becomes involved with the child due to an acute admission to hospital or one who has provided on-going specialist expertise regarding a child's feeding or breathing difficulties. However, this was variable, and in some cases a recommendation is made by a dietician, speech therapist, physiotherapist, or nurse. For others, the recommendation comes after a family has actively pursued one, having already made a decision that technological support is required.

There is variation in where the recommendation takes place too, with some families identifying the traditional consultation and others identifying a children's hospice, a hospital ward, an outpatient clinic, the home, and in some cases a telephone conversation. When recommendations are given during a consultation, some parents believe it is useful to attend with some-one else, and Charlotte's mum admits that because she is quite an emotional person, she will not always absorb the information or interpret it accurately depending on her emotions at the time. She therefore relies on her husband to process the information, which they then discuss at home.

Table 5 – Types of Information for Decision-Making

Information Type	Main Sources
The Recommendation	<ul style="list-style-type: none"> - child's paediatrician - other consultant - nurse or allied health professional - social worker
Expert Opinion	<ul style="list-style-type: none"> - child's paediatrician - other consultant - research evidence
The Risks and Complications	<ul style="list-style-type: none"> - child's paediatrician - other consultant - surgeon - nurse or allied health professional - other organisations (e.g. charities)
Test Results	<ul style="list-style-type: none"> - health service - hospital
The Procedure and Equipment	<ul style="list-style-type: none"> - child's paediatrician - other consultant - surgeon - nurse or allied health professional - other organisations (e.g. charities)
User Experience	<ul style="list-style-type: none"> - other families - healthcare professionals - other organisations (e.g. hospice, charities) - internet forums and groups
Condition-Specific Information	<ul style="list-style-type: none"> - condition-specific charities - medical professionals - research evidence - general internet resources

Both Matthew and Harry agree, and value having their parents present so that between them they will have a better understanding about the recommendation being made. Lee's mum also talked about this, and made sure that if she could not accompany Lee, a carer would go with him instead.

"You see that way you see, coz sometimes I miss things they might hear and they might miss things that I might get. So it's a way of getting more, different views."

Harry values attending consultations with his parents.

The way in which a recommendation is presented is crucial, and a distinction is drawn between a 'recommendation' and a 'suggestion'. In some cases where a recommendation is phrased as a suggestion, the professional involved lacks the authority to make an official recommendation. Therefore, for some families a subsequent and more formal recommendation will be made by the child's paediatrician or another consultant.

However, this distinction also relates to the strength of the recommendation being made, which can be seen in Molly's case, as the recommendation for Molly's gastrostomy was perceived very differently to the subsequent recommendation to change her gastrostomy and perform a fundoplication.

"Doctor C [Molly's main paediatrician] suggested a gastrostomy because she went through a really bad stage where she were just losing loads and loads of weight. And he said "I know you're doing well feeding her but it's just not enough"."

The gastrostomy recommendation for Molly was perceived as a suggestion.

"So they persuaded me to have a gastrostomy [fundoplication].....The doctor [a surgeon] were just going on and on. He was saying that instead of thinking about myself I should think about my daughter and all this. So I agreed to have it done but that's what she died from.....I felt really pressured. I were upset that I'd been pressured into changing

my mind, and I wasn't happy."

The subsequent recommendation for Molly was perceived as a command.

The perceived strength of a recommendation is therefore significant, because it has the potential to alter the perceived choice in the decision being made, and can be interpreted as the opinion of the referring professional, even when they are not explicitly offering their views. The following examples give a real flavour of the different ways in which recommendations are interpreted by families.

"I think the medication, people just expected her to take it. It was not a choice because it was a matter of when she started fitting she should take the medication. In fact yes she did have to make some choices, and in the end it was really a matter of take it but having the lowest dose."

Kate's mum talks about how recommendations can sometimes disguise choices.

"It wasn't like "you will have it done". You know, it was like "we would advise it but if you don't want it we can't actually enforce, we can't make you do it". Which is fair enough. I mean, they can't make you do something you don't want to do."

Peter's mum describes the different ways in which recommendations can be made.

"She says "right that's enough. Don't feed her no more". She picked her phone up. She says "go down onto the children's ward", and she says "she's gonna have an NG tube passed." We were like "oh my god what?""

Emily's dad describes the commanding nature of the recommendation.

Given the distinction families make between known and unknown professionals, and the importance of building partnerships and trust with the professionals who are involved in caring for a child, it is not surprising that parents can find it difficult when the recommendation comes from some-one not previously known to the family, and assessing the strength of a recommendation can be more difficult in these cases.

“If that had been a consultant on the ward I wouldn’t have taken their opinion into account. I might have listened to them and said “yeah, yeah, OK”. But I would then have just made up my own mind regardless. Whereas Doctor C, he knew his quality of life. He knew him. And he knew how much he’d already suffered.”

Lewis’s parents describe the influence of their main paediatrician.

“It’s difficult when you meet somebody for the first time. And you’re having to make decisions straight away when you’ve not had time to weigh up what they’re like. You know “do I trust this consultant or not?” Coz there’s some proper dodgy consultants. Well you see it all don’t you? When you’ve been in the NHS that long.”

Peter’s mum identifies the difficulties of making decisions with unknown professionals.

This can be seen in stark contrast to decisions involving a professional who the family already know and trust, and who knows the child well. In these cases the recommendation can be both a valuable source and type of information because the individual opinion a consultant has to offer is based on the knowledge they have acquired over time about the child and family.

“Doctor C helped us to make the decision. That was his consultant at the time. He came over to see us and he said “well these are the options. We could try this”. And I said “what do you think about that?” And he said “well my feeling would be not to.” You know, and we really trusted him you know.”

Lewis’s parents describe the overlapping nature of a recommendation and opinion.

Consequently, for some families the recommendation is the only source of information a family needs. However, for others the recommendation is the only source of information a family has access to, particularly if their child is in hospital and there is little opportunity to access information or advice elsewhere. When the professional involved is not known to the family, the information available to them can therefore be very limited.

9.4.2 Expert opinion

Expert opinion was identified by parents and young people as the most important type of information, and in the main is provided as part of the recommendation, and from other medical professionals families have access to. Although a type of information, it is in essence the 'opinion' of the medical professional it comes from, and parents and young people both identify the potential biases and agendas underpinning this information, in some cases seeking out a 'second opinion' and alternative sources of information to check for any bias or hidden agenda.

"They may be a specialist but if you're going to see a surgeon that's what they're considering you for ultimately. And I think people don't see it like that. What they see is this person who has apparently got an open agenda."

Hollie's dad believes surgeons have a bias towards surgery because of their profession.

"I mean they try and do what they think is right. And again it's their opinion. It's their opinion. They might know more about medicine, but that's not everything they need to know, because not all decisions are based around that. It's about quality of life and lots of things, not just that."

Harry talks about the need for other types of information.

Nevertheless, despite the perceptions and beliefs held by parents and young people about the biases and agendas of 'consultants' in particular, the data revealed a strong consensus among families that the information and opinion provided by the consultant is the most influential information they receive. Ultimately, families still view the consultant as the 'expert', and welcome their opinion even though their trust in the medical profession as a whole is sometimes called into question.

"Who do you listen to? The consultant I suppose, rather than somebody else. I mean they're the experts, not someone you met.....so most of it is from the consultants. And it should be. Because the trouble is you're never gonna get everybody agreeing about everything anyway, and the

danger is you can end up with information overload.”

Harry’s dad talks about the importance of medical expertise.

Families particularly welcomed having access to a second opinion, in other words a second source of medical expertise, and families with this resource described themselves as very fortunate. In most cases, the second opinion comes from their child’s paediatrician or other specialist consultant with whom they have an established relationship, and a children’s hospice doctor was also identified by some families as providing information and opinion on the use of technological support. However, this expertise is not limited to paediatricians and consultants, and parents and young people referred to the role of highly knowledgeable nurses and allied healthcare professionals who provide insightful and influential opinions regarding the on-going decisions families must make.

“I rang Doctor C, who was Molly’s paediatrician. And he said “I can’t advise you what to do”. And I said “if it were your child would you have it done?” And he said “I don’t, I can’t say.” He said “but in Molly’s case I think I would but that’s a really unofficial opinion.”

Molly’s mum respected the opinion of Molly’s main paediatrician.

Young people too talked about the importance of getting a second or even third opinion when making decisions, although Harry admits that sometimes too many opinions can make it difficult to decide, particularly where there are opposing views on the decision under consideration.

“It’s helpful but it’s also sometimes hard to decide because if two of them don’t agree you’ve got to work out which you think is best.”

Harry talks about dividing opinions among medical professionals.

However, Harry also recognises that when there are divided or varying opinions, the option of doing nothing is real, and there is more perceived ‘choice’ in the decision. This raises a concern for families who only have one medical opinion available because the perceived choice is channelled through the individual offering that opinion.

“It’s difficult but then at least you’ve got a choice to make. If you only get like one, either side, whichever side it is, then you’re just forced into whatever they make you believe, coz you’re gonna believe your consultant over anybody else coz they’re supposed to know what they’re talking about, which they don’t always, but they’re meant to.”

Harry talks about dividing opinions among medical professionals.

9.4.3 The risks and benefits

Parents identify the risks and complications associated with technological support as an important type of information for decision-making, and parents and young people in the main identify the recommending consultant and in the case of surgical interventions, the surgeon and anaesthetist as the key sources of this type of information.

“The surgeon was very, very good and he went through all the problems and made it perfectly clear to us that Sebastian might die.”

Sebastian’s dad identifies the role of the surgeon in communicating risk.

Parents welcome this information so as to weigh up the potential benefits and risks associated with the proposed intervention. However there was less consistency within the accounts of young people about the value of this information, some of whom are overwhelmed by the sheer amount of information about clinical risks and complications.

“Sometimes you think you’re better off not knowing all the risks. Sometimes I think, coz it keeps on, it makes you keep on doubting it, whether to do it. Coz some risks aren’t risks, they are just ridiculous.”

Alex would prefer less information about the risks involved.

“I find surgeons are often very keen on expressing risks. I find that a lot, they express risks a lot more than other doctors, other consultants.”

Richard identifies the surgeon as the key provider of information about risks.

The young people taking part in the study also draw attention to the technicality of the information being presented, and recommend that it is offered in a way that young people can relate to if they are to understand the potential risks and benefits involved in a proposed intervention.

“The thing with me is a lot of technical information just goes in one ear and out of the other.....but I mean, the information they gave me did help to decide you know that the benefits of it kind of outweighed the whole negatives at the time I had for it.”

Matthew sometimes finds it difficult to process the technical information about benefits and risks.

“I think as a child you want to know the impact it’ll have on your life rather than all the, you know, medical stuff. You want to know that you won’t have to come into hospital hopefully as much. As a child that’s easier to understand than somebody saying well this will increase your lung capacity from 17 to 30.”

Richard believes it is important to put medical information into a wider context.

Both young people and parents can find the information about ‘risks and benefits’ difficult to process, because it specifically relates to the contrast between positive and negative outcomes. However, unlike some young people who would prefer to have information about the significant risks only, many parents described checking that they are aware of all the risks and benefits involved. This is partly in response to the awareness parents develop as they make on-going decisions that not all the information is retained following the initial recommendation, or is misinterpreted during a consultant yet still used to inform the decision.

For some families though, it is part of a wider process to check that they are properly informed to make a decision, and where families have experienced decision processes in which the referring professional has not communicated all the risks, or where families recall this to be the case, this process of checking offers families reassurance that they are properly informed. Families identify that this type

of information is perhaps the easiest to access because it is readily available online, and can often be found on reliable websites such as those belonging condition-specific charities. Some families seek out the research evidence when considering the risks and benefits, which they explained is a means of assessing the level of risk involved.

“We actually looked at clinical trials that had been done, you know, so we knew the pros and cons.”

Harry’s dad draws on research evidence about the risks and benefits of interventions.

9.4.4 Test results

Although parents stress the importance of considering their child as a whole being, and their quality of life as a central factor in the decision, test results and medical evidence can help parents understand why their child needs a gastrostomy or ventilation and are therefore identified as a type of information. It is worth pointing out though, that families will not always be given automatic access to these results, or be able to interpret them correctly, and are often reliant on the interpretation of the person communicating the results. Nevertheless, it can provide solid evidence of a child’s difficulties, which for some families reduces the uncertainty in the decision being made.

Dad: “I mean it were, we put the video-fleuroscopy wasn’t it, and then, I think that were when the final decision to give her a gastrostomy coz they said, they looked at the video-fleuroscopy and they could see it wasn’t doing what it was supposed to.”

Mum: “You could see. I had to feed her this like barium meal. And when she swallowed you’ve got like a flap apparently. This flap comes down when you swallow and it goes into your stomach. It wasn’t doing it in time, so it were going onto her chest. So that’s why she were getting chest infections all the time.”

Emily’s parents talked about the importance of the medical tests, which helped them to see why she needed a gastrostomy.

Like Emily's parents, Sebastian's dad identified the test results as information that helped both Sebastian's parents and the professionals involved to determine whether or not Sebastian needed a gastrostomy.

"He did have some tests and they confirmed from swallowing the liquid that swallow function was malfunctioning and it would go into his lungs."

Sebastian's dad identifies the test result as a type of information for decision-making.

However, for other families who identify the test results as a type of information, they merely confirm what parents already know, and are instead viewed by parents as a type of information for the professionals instead. This was evident in the decision for Casie to have a gastrostomy, because the professionals involved did not believe she had swallowing difficulties until Casie was admitted to hospital and her parents demanded that action be taken.

"Casie became ill with pneumonia and ended up in hospital.....And she had a video-fluoroscopy and nobody needed to explain to me what was going on. It was so obvious. It just went straight down both tubes, straight down. It was 50:50 down both when she swallowed."

Casie's test results confirmed mum's opinion that Casie needed a gastrostomy.

This contrasts to Matthew's decision about assisted ventilation, in which the information generated from his overnight sleep study was instead used to determine that, despite Matthew's wider symptoms of respiratory weakness, his lung capacity was sufficient to carry on without support from a ventilator.

"The professionals were looking at the overnight SATS results and saying, well actually he's fine."

Matthew's mum believes the test results, while important, should not be decisive.

9.4.5 The procedure and equipment

Information about the procedure and the equipment a child or young person will use was identified to help parents and young people understand more about the

technology itself, and also about the potential impacts of living with the additional equipment they need. Nearly all the families taking part identified receiving this information, and although viewed as secondary to both medical expertise and user feedback, it is valued by families because of the increased understanding parents and young people reported having after receiving the information.

“We had to go into the hospital to show us the actual machines first, to see how it felt on me and stuff. And they let me try the masks on first as well when we were there, and told us the pros and cons of both different ones.”

Matthew talks about obtaining specific information about the ventilator and masks.

Some families recall receiving visual or written information, others were informed verbally by their consultant or the recommending professional. For others parents and young people, professionals organised for them to see the equipment themselves, or to meet other children and young people already using the technology so they could see first-hand what life might be like. As well as obtaining this information from professionals, families identified the internet as a resource for information about the procedure and equipment, and some were able to draw on their prior encounters with other families whose children had a gastrostomy or received ventilator support.

9.4.6 User feedback

The experience of other users was identified as one of the primary types of information, and for parents and especially young people, knowing what life will be like afterwards is crucial information they are not always offered from the referring consultant.

“They may know the medical implications and things like that but they don’t know how you feel to have a mask on and things like that.”

Harry talks about the focus among medical professions on medical factors.

For parents, seeing first-hand the benefits experienced by other children and their families is particularly helpful because it can be difficult to imagine what it might be like, especially when feelings of uncertainty and fear accompany the decision process.

“The person who’s on the ventilator understands because they’re on it. They know how they feel.”

Alex’s mum identifies the value of information from other young people.

“Seeing how they felt, and what sort of emotions they went through when they had to make a decision about ventilation. And I remember her saying to me that was the best decision she ever made. And that really stuck.....and seeing her kids and her home life as well, I think that helped as well.”

Josh’s mum visited another family during the decision about ventilation.

For Lee’s mum, seeing the drawbacks experienced by other children led her to question the appropriateness of artificial nutrition for Lee, and enabled her to pursue it in a way that maximised the benefits, opting to feed him artificially at meal times rather than during the night.

“The other boy, he had an NG tube and she used to feed him overnight. Well he never, ever felt hungry. So he never ate anything through the day. And then, I’m not being funny, then he went to bed at 8 and then started the bloody process all over again.”

Lee’s mum drew on the experience of another child who was tube fed.

Young people also draw on the experience of others to understand the potential benefits of living with a gastrostomy or ventilation, and Matthew, who was reluctant to consider ventilation at first because of the invasive nature of the mask, was able to use this information to minimise his fears and see the benefits despite the initial period of discomfort.

“I talked to one of the lads. He was on it, and I obviously asked him about, asked them about how they coped with it.....and he said “oh yeah it’s a bit uncomfortable at first but you do get used to it, quite quickly” and stuff.....it was like someone who experiences it on the same level rather than someone that might just say, try it once, in the training, it’s actually someone who uses it on a daily basis.”

Matthew found it helpful to talk to another young person on ventilation.

Harry recalls how important this information was in the decision he made, and visiting two young men who had opted for a tracheostomy was really useful and influenced the choice he made.

“We went to their house and they said we don’t get in hospital ever. Just for check-ups but everything else is done at home and all that sort of thing. And it made me think that even if you do it, you can still have a good life. You’re not stuck doing less things, because I think I thought that with a tracheostomy and a suction machine and things like that, you’re thinking about that it’s going to influence what you can do.”

Harry recalls meeting two men with a tracheostomy.

User feedback is sourced through several channels, with some families proactively seeking out online discussion forums and the websites of condition-specific charities, or making contact with families they have met along the way. Professionals were also identified as a resource, and some professionals organised for families to meet for this purpose. However, this was not the case for all families, who instead turned to alternative sources such as their child’s school, a children’s hospice, a respite provider, or by directly contacting a charity they felt might be able to help.

“There’s a charity that is run by people with my condition. And I often, if I have a big decision I will sometimes ask them if I could talk to somebody with my condition.”

Richard identifies a charity as a good source of information.

“We’ve talked to parents in the [condition specific charity] group. And again it’s useful because there in the group you’ve got parents who will have to consider it, parents who are making the decision, and parents who’ve made the decision.”

Sebastian’s dad identifies other parents as a source of information.

Obtaining user feedback is not always straight forward, and not all the families in the study had access to this type of information. Among those who did, the information was not always as helpful as it could have been, and Lee’s mum observed that many of the young people around Lee had quite severe learning disabilities which made it difficult to obtain any meaningful feedback and user experience.

“It was very difficult because the ones that he had around him, he got on with but perhaps were, this sounds bad, but they had more of a learning disability, so he didn’t really benefit from talking to them.”

Lee’s mum found it difficult to get Lee some meaningful feedback from other users.

9.4.7 Condition-specific information

Young people with well understood conditions are sometimes aware that they will need ventilation or other interventions such as spinal rods before they are recommended. Sometimes this information comes from their parents, or the medical professionals involved in monitoring their condition. In other instances young people’s encounters with other young people living with their condition provide them with the knowledge that they will one day have breathing or swallowing difficulties serious enough to warrant medical intervention.

The knowledge Kate had about her condition in advance enabled her to make a decision about technological support two years before a gastrostomy was recommended to enable her to be fed and medicated. Like other young people in the study, it was still made on the basis of what her quality of life would be like at the time a recommendation came, and the potential impact a gastrostomy would have. However, knowing that she would gradually lose the ability to communicate

with others, and would most certainly not be able to communicate at the time a decision might be required, meant that Kate was able to make informed decisions about technological support at a time she was able to do so.

The progression or deterioration in a child's condition is an important decision factor, and the evidence base regarding a child's illness can be helpful to parents in determining their child's needs. This knowledge can also be used to consider whether a child or young person is being considered for technological support at the right time.

"I was in a position when I was talking to parents who come from different parts of the country where boys were being put on it much earlier as a preventive rather than waiting for when they'd gone into breathing difficulties and then put them on it."

Matthew's mum used her knowledge about the management of Matthew's condition to consider when he should begin ventilation.

One of the drawbacks identified by families regarding the use of evidence about certain conditions is that the need for technological support can sometimes be assessed on aggregate data regarding a child's condition, or on the expected progress of deterioration. As a result, the individual nature of progression and the overall wellbeing of a child can be overlooked.

"It varies for all of them. How it takes, the illness, how it takes them varies. You can't say at 14 you will do this and at 15 this will happen, at 16 this, coz it's just so different."

Peter's mum describes how the same illness affects young people differently.

"Sometimes they put you all in the same kind of basket instead of, or because you've got a certain condition you must have these problems.....and you're like, well no.....my condition may be more severe in certain areas and less severe in others than similar people with my condition. It affects us all in different ways."

Richard talks about the danger of using general assumptions about his condition to assess his need for technological support.

In other cases, parents recall assumptions that are made about children with certain conditions, or condition types. For example, Steven's dad recalls being told by a specialist consultant that for children like his son, it is acceptable to be underweight. When Steven was then referred for artificial nutrition due to his underweight state, Steven's parents were confused about what Steven needed, and about whose advice to base their decision upon.

"He's supposed to be one of the biggest consultants for these types of children in England. And he said do not get weight on this child whatever you do, don't let anybody persuade you to overfeed him."

Steven's dad about the former advice he was given regarding weight gain.

Parents report sometimes being unclear whether such assumptions are based on the available evidence about a child's condition, and it can be difficult for them to ascertain whether this is the case or not. This point draws attention again to the way parents believe some professionals use aggregate data when assessing the needs of children with a poorly understood or undiagnosed condition, and in fact parents emphasise how essential it is to consider the needs of an individual child as a whole being, and not one simply defined by her condition or symptoms.

9.4.8 Asking questions

As a result of acquiring information for decision-making, families may find that there are certain facts that are difficult to understand, pieces of information that cannot be recalled with any specificity, and new questions that parents or young people believe need answering before they make a decision. When asked about the use of information for decision-making, both parents and young people consistently emphasise the importance of asking questions during the decision process. This was described by some families as a means to redress the information imbalance between the consultant and themselves regarding the intervention being proposed and partly as a way to check that they are informed enough to make a decision.

What's more, there is a feeling among some families that certain information is withheld by medical professionals during the decision process, and by accessing alternative sources of information and asking questions, this information may be drawn to the surface.

"They actually do know more than they're telling you.....but if you want to know something, ask them. Otherwise you're not going to know."

Harry is aware that information is sometimes withheld.

Charlotte's mum, who like Harry believes that not all the information is always forthcoming, stresses the importance of asking questions, and will also write them down before the meeting. She is not alone, and this strategy was employed by other families too.

"If you want to know something, ask them. Otherwise you're not going to know. And the other thing is if you don't write it down before you go in, a lot of the time you forget. And then you think, oh why didn't I ask that question?"

Harry talks about the importance of using written questions during a consultation.

Like parents, young people value the opportunity to ask questions in order to make sure they have the information needed to form an opinion, and to ensure they understand what is being said. In addition, young people will often ask their parents following a consultation to ensure they understand the information provided. Richard believes it is also important to ask other young people questions that only they can answer, because they have lived with the intervention being proposed.

"I would definitely say it's useful to ask people who have experienced it. Don't be frightened to ask questions. I think some people will just answer anything.....And if they do answer it well, it's something off your mind really."

Richard values the opportunity to ask young people questions about interventions.

However, asking questions is not always easy. The consultation can be short and at times dominated by the consultant. Some families report feeling overwhelmed and receiving more information than they are able to process. Other families admit it can be difficult to know what questions to ask when you are not yet fully informed, and believe that questions will often arise during the time after a recommendation is made. Having the opportunity to discuss the decision more than once and to ask questions is therefore essential. Finally, the journey parents and young people take as decision-makers can influence how confident they are at the time of a decision to ask questions, and to challenge the information provided.

“It’s made me ask a lot more. I’m not frightened of saying what I think anymore. I used to sort of think I used to look at doctors as like little gods you know, that you just listen to what they have to say and you’re a bit frightened of asking questions, but I don’t now I ask.”

Matthew’s mum recognises her growing assertiveness in decision-making.

9.4.9 Using and interpreting information

With enough time and resources, families can amass a great deal of information and opinion about the potential benefits and drawbacks of initiating technological support. Certain types of information, namely medical opinion and user feedback, are viewed by parents and young people as more influential than others and the combination of these is felt to offer parents and young people a more balanced view of the potential benefits and drawbacks to having a gastrostomy or using a ventilator.

Young people and parents describe a process of assessing the validity and usefulness of the different types and sources of information they are able to acquire, which are then used in combination to make a choice.

“You’ve still got to make a choice in the end. You’ve still got to make your, which opinion you think is best. You have to work out, you have to trust yourself to make the right choice which is difficult, but you need

to.”

Harry talks about using the information to make your own choice.

In particular, the reliability of different sources is considered and some of the views and beliefs families express about the agendas and biases held by professionals are included when parents and young people assess the information provided.

“It always helps asking, you know, people who use it themselves....The medical side of it you always kind of felt they was pushing towards it, you know sort of like it wasn’t as if they’re really thinking about the person. It was more seeing your health aspect of it.....whereas if you could chat to someone who can see the bigger picture, who either knows you quite well, or people that will actually personally use the information for you.”

Matthew believes that the information provided by consultants is medically biased.

Simon, who also referred to the hidden agendas and biases of professionals, believes that sometimes professionals go down a certain pathway for young people with his condition, recommending the same treatments and interventions at similar points, rather than treating everyone as individuals with their own unique symptoms and illness progression. This has led Simon to suspect that at times, the advice and information offered by professionals are based on their own preferences for treatment.

Nevertheless, Simon still values the role of medical opinion in decision-making, and very much views those who provide it as the experts in his care. This view was shared by all the families, who agreed that medical expertise is an essential type of information. However, obtaining the medical opinion of more than one professional was identified as crucial by many parents and young people, and again is seen to offer an insight into how complete the information provided by the referring professional is.

“You also want that extra information from consultants you don’t know, people that don’t know you, that are just generally discussing it, about, just generally discussing options about these things rather than just people that know the person because they’ve got, like I said, a sort of biased opinion about what they think is best.”

Harry talks about seeking out other sources of medical expertise to assess how biased the information provided by his consultant is.

“Our end decision would have still be mine. But it helped having somebody else’s input, other people’s decisions coming in as well.....You know their points of view came in as well and I thought yes. You know sometimes when you’re in that situation you can’t think, you can’t always think straight really can you, sort of think what’s best, and so having other people’s decisions made it slightly easier for me .”

Josh’s mum really valued the opinions of the doctors and nurses around her.

Written information about the procedure, or supplementary information found on the internet or provided by third party organisations such as condition specific charities is useful but it does not make up for the invaluable information and opinion provided during discussions with healthcare professionals, and with other families themselves. Sometimes the purpose of drawing on various sources for the same type of information is again to assess the neutrality and reliability of the information being provided by medical professionals. However, some families value having information to take away, as it supplements what is already provided by the professional involved, and can be used to work out questions to ask during the next consultation.

There was also some evidence in the data that the information families require can vary depending on the nature of the decision under consideration, and Richard believes that for decisions in which the ‘need’ for medical intervention is very clear he requires less information because he has already accepted that the choice of doing nothing is not viable. In contrast, Richard believes that where there is less

certainty about the need for technological support, more information is required in order to assess whether it is appropriate or not.

“I haven’t been ill for quite a number of years now and I think if somebody came now and said “oh you need a gastrostomy”, I’d be a bit like “well why do I need one?” And I’d question it a lot more because I’m thin yes, but not desperate, you know I’m not starving.”

Richard feels that the amount of information needed is different for each decision.

The internet as a source of information is used variably by families, some of whom describe it as a key resource for various different types of information, and others who use it to supplement and check the authenticity and reliability of the information already provided by professionals. However, there were families in the sample who actively chose not to access information online because they view it as inferior to that which is available from other sources.

“Try to find out as much as you can. You want to actually get as broad a medical advice as you can. Not, I mean the internet is fine but it’s not the same as going to see somebody and talking to them about cases in detail.”

Harry’s dad ranks the different types of information.

“I don’t know what really to trust on the internet.”

Richard is wary of information available online.

“I think I would, I don’t know too much, I wouldn’t know too much about going on the internet because I think not all sites are as good as they could be.”

Lee’s mum voices her concerns about the quality of information online.

9.5 The Health Care System

These on-going interactions between families and professionals as they exchange information and opinion during the decision process takes place within the wider healthcare system, which families also encounter at different times when their child

is diagnosed, unwell, monitored, or when their child requires therapy and additional support or care. It is not surprising then that many families identified the influence of the system as a whole on their ability to make informed decisions, and in particular draw attention to the wider constraints placed upon professionals as they endeavour to act in the best interests of the children they serve.

The on-going interactions families have with a system that is seen as fragmented and ineffective, can also influence the attitudes parents and young people have towards the NHS and those who work in it, which in turn can shape the way in which recommendations are perceived, and how information for decision-making is understood and interpreted. Three main aspects of the system were identified as having an impact on decision-making – the experience of staying in hospital; the bureaucracy and red tape; and the transition to adult services.

9.5.1 Hospital stays

Families' accounts are scattered with the trials and tribulations of staying in hospital, and the impact of these experiences is seen to have an accumulative effect on parents, many of whom over time refuse to leave their child alone on a hospital ward, or to place their trust in the medical professionals they are not familiar with. Many accounts relate to errors and mistakes that have happened, which can be distressing for both parents and children, and can sometimes result in a life-threatening situation that would otherwise have been avoidable.

“The only one time we didn’t stay, she just, she lost a day of her life because somebody messed up with drugs.....So she’s never been left after that. I mean cock ups have happened since, with the drugs, I mean they’re strong drugs....that’s why whenever she goes in now, we always say “we’ll medicate Emily”.”

Emily’s parents continue to care for Emily around the clock during hospital stays.

Some parents expressed a concern that children are not monitored closely enough during hospital stays, and that their safety is at risk unless parents remain at their bedside.

“We would be with him all the time in hospital. We wouldn’t feel like we could leave him. And then like you’d go to get something to eat or go to the toilet or whatever and you’d come back and he was fitting, and nobody had noticed, and you know, bits like that, that are just to do with it being, them being understaffed basically.”

Lewis’s parents identify their continued role as carers during hospital stays.

In the same way that families assess the experience of their child’s paediatrician or consultant in caring for children with life-limiting conditions, parents become aware of the differences on the hospital ward too, and believe that some of the nurses and doctors who work in the hospitals where they have stayed sometimes lack the knowledge and expertise to care for their child.

“He was bleeding through the trachee and it was because, we don’t know, but a trachee was put in that shouldn’t have been. He had a size six in and they put a size five in so Alex was bleeding. I mean I didn’t know that Alex had a size five in until we got to hospital and they were going “we haven’t got a clue about him”. They didn’t know nothing about him.”

Alex’s mum draws attention to the lack of knowledge about Alex’s condition in hospital.

“There was an episode when we were in hospital where, sadly, they were basically killing Sebastian.....And I said “look, there’s the medication being given, and two hours later the problems start”....And eventually the consultant agreed to stop the medication.....And they, there’s lots of horrendous stories about our experience in mainstream hospital.”

Sebastian’s dad recalls one of the many negative experiences in hospital.

Some parents learn to accept that many of the professionals they come across will not know how to care for their child, recognising the complex and individual care they themselves provide, and in the knowledge that their child has a rare condition that many professionals may not have encountered before. However, the expectation parents have for their child’s paediatrician and other consultants to

acknowledge the expertise of parents is extended to hospital staff, who parents believe should do the same, especially if they lack the skills to perform some of the technical tasks associated with a child's care. Yet many parents report this not to be the case.

"She choked from the reflux several times a day and during the night, particularly during the night because she was laid down. And none of the staff ever saw it and they didn't believe that it was happening. And she was on a SATS machine the whole time. And you knew when it was happening because her SATS would drop, but there was no outward sign that she was struggling because it was really subtle. And everybody else claimed it wasn't happening."

Casie's mum describes the frustration of not being listened to by the ward staff.

Not all hospital stays are quite so eventful, and there were some accounts within parents' narratives in which the steps put in place to ensure children receive the care they need when they are admitted to hospital work well. Emily's parents described the system in place at their local hospital which enables them to go straight to the children's ward when Emily requires medical attention, adding that the ward staff know her really well now because of the relationship that has been established over many years.

Nonetheless it was clear from the accounts of parents, and especially young people that they will do whatever it takes to stay out of hospital, and while these on-going experiences are not directly related to decisions about technological support, they nevertheless influence the way in which procedures that require a hospital stay are viewed. They also have an impact on the evolving parent professional relationships, and the commitment parents have to ensure their child is not left alone during a hospital stay can cause friction between parents and the ward staff, who parents believe sometimes view them as neurotic and overprotective.

"There's just a few people that I find, no matter which way you go with them, you can't win. You know, you're an over-protective mother. Well I

tell you what I'd rather be an over-protective mother than not care about him at all."

Alex's mum describes the attitude of some professionals towards her.

"Well we were labelled as neurotic parents weren't we? We were labelled as completely neurotic. But that's something you just have to put up with."

Casie's mum believes that some professionals labelled them as neurotic.

9.5.2 Bureaucracy and red tape

From dealing with professionals over a sustained period of time, and sometimes staying in hospital on a regular basis, families become aware of the rules and bureaucracy within the healthcare system that can sometimes prevent things from happening, cause delays, and require families to 'battle' with services and professionals to ensure their child receives the care and support they require. For many families, the system is viewed as fragmented, difficult to access, and unable to cater well for children who will require support for a number of years, and from various services and professionals.

"These organisations are like super tankers.....They've got a momentum of their own and they don't, they're inflexible. They can't cater for special circumstance. They're sort of like sausage machines. So children like Sebastian, they can't cope with."

Sebastian's dad describes the healthcare system as inflexible.

Many parents report barriers in accessing support and equipment when it is needed due to the unnecessary rules and red tape, and Casie's dad highlights the problems of trying to speak to a consultant when the system is one of appointments and bookings.

"I phoned the consultant and I was told by the secretary of one consultant "consultant's don't talk to the patients". You know, you can't ring up and have an informal conversation. So when you're meeting

barriers like that it's difficult."

Casie's dad identifies the bureaucracy that makes it difficult to obtain advice.

"What a nightmare getting his wheelchair to his size for when he had his back done. It's got to be specialised done, and somebody had cocked it up somewhere along the line.....so they wouldn't really put him in his wheelchair would they? But nobody would sanction to do it. And you just think it's bloody ridiculous red-tape. Time and again, it does your head in."

Sam's dad describes the red-tape that can prevent things from happening.

Professionals are crucial here, and where families are surrounded by a team who will do what it takes, they are often seen to bend the rules and red tape that can stand in a family's way to getting the help they need.

"There's the people at the bedside, and they know what is happening. And in a way they support and keep going what is very, a very insensitive system. Full of rules and bureaucracy. And they put a human face to it, and they often bend the rules in order to make the system work.....If people followed the rule and just did the job then the quality of service is appalling."

Sebastian's dad identifies the role of professionals in helping to navigate the system.

Some families learn how they too can navigate the system to their advantage, and describe the strategies they employ to get their child the care, equipment, and support they need.

"Until we learnt how the system doesn't work, and how you can get round it, other parents, and I've heard this time and time again from other parents, who don't realise that you can bypass so many things just by asking different people to help you out."

Casie's mum learned how to navigate the system by utilizing key professionals.

Like the experience of hospital stays, the barriers and bureaucracy associated with the healthcare system influence the developing relationships between parents and professionals, and the views of parents about the NHS, and about those who work in the system. When barriers are seen to prevent children from accessing the support they need, parents can become angry and frustrated. These influences are important because of the crucial role of professionals in enabling families to make informed decisions, and the part played by professionals and services in providing information and support following the initiation of technological support, as well as for the decision itself.

9.5.3 The transition to adult services

Within the data collected from families who have experienced the transition from child to adult services, it is evident that the team of professionals surrounding a young person can change quite dramatically once their care is provided by the adult system. The age of transition is variable, and for some young people the changeover can be very sudden when services have specific cut offs such as sixteen or eighteen. For others the transition is more gradual, with some services identified as having a more flexible approach. Because families are often being supported by numerous services, transition can also vary by individual organisation and some families continue to have informal access to key professionals in paediatrics, whereas for others the transition is final and all ties are severed.

“When it’s children, to eighteen they’re good. But after that you’re on your own. After they’re eighteen you’re on your own. You’re struggling.”

Steven’s mum identifies the lack of support available in the adult system.

“There was just nobody when he got to a certain age. I said “well who does he transfer to?”. “Well we haven’t really got anybody who’s a specialist in that field”....but we still went to the children’s hospital, even though Peter was you know an adult. They don’t seem to demark it as much there.”

Peter’s mum identified the limited services for young adults.

Overall, the transition is not regarded as a positive experience because of the loss of trusted professionals who families have spent many years building relationships with, and who have an in-depth knowledge of the young person and their family. Where key decisions are made around this time, the resources families have available to them are reduced, and in some cases simply not available.

“They’re passed off from the paediatrics and that’s the problem. Things afterwards get missed because they get to an age and they’re taken away from, they should have someone.....It’s like they get to an age and it’s like they’re no longer noticed. They’re forgotten about. And that’s when something goes wrong.”

Alex’s mum describes the problems with the transition to adult services.

“Well it’s like you’ve just been forgotten really, at a time when his needs are increasing. You know, it seems like the actual support, health-wise, is less.”

Matthew’s mum believes that there is less support after the transition to adult services.

As well as the loss of key resources for decision-making, families talk about the separation of services due to the different infrastructure making up adult health care. Some young people will have several new consultants, none of whom take overall responsibility for their care, and none of whom specialise in their condition, instead each having an organ or function which they accept responsibility for. In some cases, these professionals can be based at different hospitals therefore further fragmenting the care young adults receive. For families who are able to maintain a relationship with one of the professionals they have come to trust over many years, this resource is highly valued, and can become the primary contact for on-going problems their adult child may now experience.

“I mean Doctor [child consultant], he’s our life-line. Because when they get to Matthew’s age everything’s fragmented off with different doctors. So it’s always Doctor [child consultant], he’s the first person I ring and

then he'll know who to contact."

Matthew's mum identifies the key role of the consultant who has stayed in contact.

Holding onto this resource is important to families following the transition to adult services, and this is reinforced by the lack of knowledge families identify the newly assigned professionals to have about the specific condition their child has, especially when compared to the expertise and knowledge held by the specialist consultants and nurses involved in their child's care up to that point.

"Well I don't think many people to this day know a lot about the condition, but especially when you go into adult, you just lose all that. I mean, down to the consultants or the respiratory nurse. Nobody actually understands."

Alex's mum identifies the loss of expertise following the transition to adult services.

"Unfortunately there's a team there [local hospital] that's been fantastic and I mean fantastic but unbelievably, with him being an adult you see....they're all, they were children's, and unfortunately this new speech therapist didn't know the score you see."

Steven's dad compares the children and adult services involved in Steven's care.

Finally, the transition to adult services was identified to have an impact on the decision-making dynamics between parents, young people, and professionals because of the assumed patient-professional dyad within the adult system. Consequently, the evolving role parents assume as decision-makers for their child, and the expertise they acquire over many years regarding their child's individual condition, is sometimes not taken into account by professionals. For Alex's mum, her continual involvement in the process of decision-making around Alex's gastrostomy led to conflict between her and the consultant involved, and she described being excluding from the decisions being made, despite the shared approach they have always applied.

"I think Alex needs to know that whatever decision he makes we all support him on that. But being a mum, if I think something's wrong and

you feel it, you have to say it. But a few [referring to professionals] don't like it, they go "mum's got a big influence". But then I will have because I've looked after him for 22 years.....and Doctor G at one point [adult consultant], at the very beginning he would not go and see Alex unless I'd left the room."

Alex's mum identifies the conflict between her and Alex's adult consultant.

Matthew's mum also referred to the different way in which professionals in the adult system viewed her role in decision-making.

"Now that he's in adult services they talk to Matthew anyway. Sort of some of them look as if to say "well why is his mum with you", you know. Sometimes I feel like I shouldn't be there but I have to be. I mean I've never had it said, you know, you stay out of the room, but I once remember the first time we went to the chest clinic and they called his name, and I were following him on, and she went "oh?". I said "I'm his mum", and she went "oh mum's coming too"."

Matthew's mum feels like her role is questioned by adult professionals.

This shifting dynamic was recognised by young people in the study although for most their experience of decision-making continues to be one of a shared process with their parents. In addition, despite the difference between the two systems, and what this can mean for the decision-making dynamic between young people and their parents, in Richard's experience he is still viewed by some professionals as lacking capacity due to his disabilities.

"In terms of the consultants and nurses, there was certainly a big difference between adults and paediatrics. But in terms of my family it was more gradual.....But in a lot of other places some professionals believe that you can't make the decision, which is quite wrong. Even now some of them look at my carers before they look at me."

Richard's experience of both systems enables him to recognise the different dynamics.

9.6 Chapter Summary

The themes presented in this chapter describe the factors in the model that are in the main beyond the control of families because they relate to the healthcare system with which families must engage in order that their child receives the support and care they need. This category is particularly important because in practice terms, steps can be taken to ensure that the process factors act as enablers rather than constraints on the process of decision-making families engage in. This in turn can have a positive impact on the lives of children and young people with life-limited conditions and their families because of the interactional relationship between decision features (the family) and process factors (the healthcare system).

The final chapter will position the findings and model within the existing literature base, focusing on the ecological nature of treatment decision-making and the way in which the combination of decision factors, decision features, and process factors influence the perceived choice families have. In considering policy and practice implications, specific attention is paid to the process factors presented in this chapter and the information needs of families for decision-making as identified from this research. The way in which the model portrays how the range of factors influence perceived choice is also discussed with reference to the continued focus in policy on informed choice in healthcare.

- CHAPTER 10 -

EXPLORING THE DECISION PROCESS

This final chapter discusses the findings from the study with reference to the wider literature base. It focuses on the ecology of treatment decision-making in the model, and the interaction of decision features and process factors that happen around the on-going decisions that are made for children and young people with life-limiting conditions. Although the decision outcomes and consequences are not the focus of this research, the chapter considers the wider impact of the process factors identified, which are seen to both enable and constrain families as they make decisions, and affect the perceived choice and decision conflict experienced. This chapter also considers what the research tells us about the models of treatment decision-making presented in Chapter Three before setting out the study's contribution to knowledge, the strengths and weakness of the study, and the areas identified for further research. Finally, the chapter brings the research to a close by considering some of the policy and practice implications arising from the study.

10.1 The Ecology of Treatment Decision-Making

This research encapsulates the inter-related yet distinctive components that make up the overall process of treatment decision-making. By examining the process rather than the decision, the ecological model developed from the research portrays how the combination of decision factors, decision features, and process factors can influence the choices families believe they have when making decisions for their child. The model excludes from it the decision that is reached by families, and focuses instead on the perceived choice families have when making decisions, which will ultimately influence the decision they reach yet may not necessarily reflect the choice they believe they have. The model also portrays how the on-going interaction of decision features and process factors can alter decision-making dynamics over time, both enabling and constraining a family's ability to make

informed decisions, which is central to the process of deciding, even when there is no apparent choice to make.

One of the key features of the model is the ecological relationship between what are essentially two separate entities, the family and the healthcare system, which interact throughout a child's life to minimise the impact of a child's life-limiting condition on their quality of life. While the interaction of these systems does not directly influence the outcomes of a decision, it does have an impact on the way in which the decision process is experienced, and the perceived choice in the decision families are asked to make. Moreover, each decision experience and the process factors that affect it can then influence the way in which families make subsequent decisions, both in their role as decision-maker, and due to the influence the decision experience can have on how families interact with professionals and services involved in subsequent decisions.

This research therefore offers a new way in which treatment decision-making can be understood, by combining key features about the patient population, in this case families of children and young people with life-limiting conditions; and the system with which they have to engage with in order for their child's needs to be met, which for treatment decision-making in England is the National Health Service (NHS). Rather than viewing the decision at the micro-level, in other words one that happens between the patient and healthcare professional, which is often assumed to be the case in the conceptual literature concerned with treatment decision-making (Charles et al., 1999), the new model considers the whole system thereby taking into account the wider factors situated around the decision process, and the on-going interaction between the family and the NHS which continue to influence the factors identified.

Viewing the family as a system then, there are features that influence both the process of weighing up the use of technological support, and features that affect the degree to which families take part in the decision process. These features are not fixed, and change over time as a child's condition progresses and as families

make on-going decisions. Some of the features identified in the model are discussed in the wider literature concerning life-limited children and young people. For example, the evolving role of parents as they assume the responsibility of caring for their child's health care needs both influences and mirrors to some extent the evolving role of parents as decision-makers identified in the model (Kirk and Glendinning, 2004; Thorne and Robinson, 1987), with parents moving towards a position of control and expertise, which in turn enables them to take responsibility over the decisions that must be made for their child.

Similarly, the child's developing capacity for decision-making is discussed in the wider literature (Moore and Kirk, 2010), and although the extent to which children can be involved in the decision process is context and decision specific, it is an important feature of paediatric decision-making, and one that has received a great deal of attention within policy and practice (DH, 2004). Like the literature concerned with treatment decision-making for adult patients, there is an assumption that children and young people should be encouraged to participate to the extent their capacity allows (Alderson, 2007). There is also a growing body of evidence showing that young people wish to be involved in the decision process (Moore and Kirk, 2010), and the findings from the study reported here support this, with young people providing clear accounts of their role in decision-making, and their desire to make decisions for themselves.

However, the study also reveals the continued role of parents as young people assume control of decision-making, and young people value the expert knowledge their parents have regarding their condition. The study draws attention to the way in which young people utilise their parents for information and advice, even when they make independent choices as adult patients, and this mirrors the findings in recent research about young people with life-limiting conditions, which also identifies decisions as shared between young people and their parents (Bluebond-Langner et al., 2010; Mitchell et al., 2011). Viewing the family as a unit therefore brings out the dynamics between parents and young people, and represents both the developing capacity of young people for decision-making and the continued

dependence among young people with life-limiting conditions on their parents due to the progressive and complex nature of their conditions. Consequently, the model avoids the assumption implied in some of the literature that young people, once they have capacity to make decisions, no longer require the views or input of their parents (Baston, 2008).

In developing the model, the research considered the ecological nature of the relationship between the two systems in decision-making with reference to existing decision-making theory. Although there is a general approach within this to focus on the individual interaction between the patient and physician, the conceptual literature often refers to the earlier work of Parsons (1951) when describing the shift from medical paternalism towards shared and informed decision-making, describing his account of modern medical practice as an example of the paternalistic doctor-patient relationship. In this, Parsons (1951) describes the doctor, due to his/her technical competence and affective neutrality, as the 'professional' in the relationship, and the patient, who adopts the 'sick role', is admitted under the care and control of the trained professional.

Influenced by his position as a structural functionalist, Parsons' argues that because "participation in the social system is always potentially relevant to the state of illness, to its etiology and to the conditions of successful therapy" (p.431), the practice of modern medicine becomes a distinct sub-system of society, or as defined by Parsons, "a 'mechanism' in the social system for coping with the illnesses of its members" (p.432). Parsons therefore links the doctor-patient relationship at the micro level to macro-level social structures, proposing it to be a social as well as individual relationship. Consequently, the 'professional role' of the clinician and the 'sick role' of the patient become institutionalized, embedded in society, both at the macro- and micro-levels. The research reported here reveals the continuing influence of those embedded roles as parents and young people describe the on-going encounters with medical professionals, and their shifting beliefs and opinions about their respective roles in decision-making.

Positioning decision processes at the macro-level therefore incorporates both the developing relationship between families and healthcare professionals, and the wider influence of the system as a whole. Situated on the opposite side of the decision 'black box' to the family, the healthcare system and the factors and structures within this that influence decision-making incorporate within the model the wider context of the decision process. This offers a unique approach to decision-making because the role of professionals in decision-making is no longer viewed at the micro-level.

Instead, professionals can be seen in relation to the wider system in which they work, drawing attention to the wider influences of the structures, philosophy and culture of the NHS, and how these might enable and constrain health care professionals as they support families in decision-making. By viewing the decision as one that takes place within a wider structure, the model also identifies how the on-going interactions families have with other professionals and services can influence individual decisions, and affect the way in which families view the role and opinions of healthcare professionals, and the information they are able to access for decision-making.

10.2 The Consequences of the Decision Process

“Just because the decision might have been wrong because it didn’t work out, it doesn’t mean that the process was wrong, or that we made the wrong decision.”

The above depiction of decision-making offered by one of the young adult’s taking part in this research encapsulates the different ways in which the outcomes of a decision can be understood, and this is discussed by Connelly and Reb (2005) who distinguish between outcome regret, option regret, and process regret, all of which were identified by families in this study as they described their role in decision-making more generally. Therefore, although this research focuses on the process of decision-making, paying attention to outcomes is important because they offer an insight into how each decision process alters the dynamics for subsequent

decisions, and equally how subsequent decisions alter how families reflect on the original decision.

Firstly then, the outcomes of a decision shape how the decision process is viewed by families, who often reflect on the process with reference to the actual benefits and drawbacks of the decision made. Whilst the outcomes are not the focus of this study, it is important to acknowledge their influence on families' narratives, and Harry himself observed this, explaining that if the research had taken place six months after his decision to have a tracheostomy, his narrative may well have been very different because of the difficulties him and his family experienced during that time. Not only is there a period of adaptation during which children may not initially experience the expected health benefits, but the additional equipment required to administer technological support can have an impact on their everyday life and their ability to take part in the activities they were able to do before, a point discussed at length by Heaton et al. (2003).

In fact, the impact of decisions can be far-reaching and difficult to predict, and many families described the subsequent and on-going decisions they have made regarding the technological support being used. Some children also experience complications and side effects, requiring the use of other equipment, or the need for additional and sometimes surgical procedures, a finding already highlighted in the existing research concerning the outcomes of gastrostomy and assisted ventilation reviewed in Chapter Two. For some families then, the decision about a gastrostomy or ventilation is one among many they will make over a period of years about their child's feeding or breathing problems, each having the potential to alter a family's experience and subsequent narrative regarding the original decision they made.

Furthermore, the need for technological support may arise from a medical crisis at a time when their child has been admitted to hospital, or it may have been discussed for a number of years as parents plan for the future. It is also a decision that comes following many other decisions parents have made for their child regarding their

care and treatment, and one that will be followed by other, complex decisions they must make in the future. The unique role parents play over the months or years of their child's illness – as their child's guardian, their advocate, and their carer – means they will experience many encounters with a range of health care and other professionals, both positive and negative, which will each alter the dynamic of the decision-making context, and influence the role parents, children, and health care professionals play in subsequent decisions for their child.

The outcomes above relate to the impact of the decision that is actually made. However some of the outcomes a family can experience are a direct consequence of the decision process. The combination of decision features and process factors, which interact over time and alter the decision dynamics between families and professionals, offer an explanation as to why this can happen. In particular, the range of professionals families have access to, the stage at which relationships between professionals and parents have developed, the time and information for decision-making, and the prior encounters and events leading up to the decision directly influence the experience of making decisions. Moreover, the on-going encounters families experience with the healthcare system, and their journey to becoming informed and expert parents, also have the potential to influence the decision process, affecting how the recommendation is interpreted, and shaping a family's understanding of quality of life, of their values and beliefs, and the needs they might have for information and medical opinion.

For parents, the consequences of a poor decision process can be significant, leading to feelings of anger, regret, remorse and distress, both towards themselves as the individuals with decisional responsibility for their child, and towards the professionals involved in the decision process. Similarly, where the factors around the process enable families to make an informed choice, this can be experienced positively, and in the same way that a poor experience can reinforce the feelings of anger and frustration towards the healthcare system, a positive decision experience can break down these feelings, and challenge the negative beliefs that families

might have, and for some families enhance the positive relationships they have already established with the professionals involved in supporting their child.

10.3 Exploring Decision Conflict

The literature concerned with making complex decisions which involve uncertainty and require families to weigh up the benefits and drawbacks with reference to quality and prolongation of life, consistently draws attention to the potential conflict in decision-making that may occur between a child or young person and her parents; between two parents; between parents and professionals, and among professionals due to differing opinions about what constitutes the right decision (Whitty-Rogers et al., 2009; Wright et al., 2009). However, as Wright et al. (2009) point out, conflict is relatively rare, and in most cases can be managed by those already involved in the decision-making process.

The findings from this study support this assertion, and interpersonal conflict about the decision itself was rarely reported. In fact, while both parents and young people spoke in detail about the process of deliberation, the differing of opinions and values of professionals, the potential biases in the recommendations being made, and in some cases disagreements about which course of action to follow, it is not something that parents find unusual, and is instead viewed as an expected element of the process, particularly when there is no clear path to take and a multitude of benefits and drawbacks. Having said that, conflict does arise between families and professionals, but rather than being related to the ethical and moral dilemmas that are assumed to surround these decisions, it is in most cases due to elements of the decision process that constrain families to make the right choices for their child.

The difficulties that can exist in the evolving relationships between parents and professionals are a case in point here, because where families have access to trusted professionals who have taken the time to get to know a child, who acknowledge the growing expertise and knowledge of parents, and who act as an accessible source of information and opinion, the relationship of mutual trust and

respect that is established can cope with the differing opinions that may occur regarding the on-going and sometimes difficult decisions that have to be made. However, when families are asked to make decisions without the invaluable resource that professionals can become, disagreements have the potential to cause conflict in a relationship that is neither established nor respected.

Sometimes, quality of life *is* at the centre of these disagreements and potential conflict, but the evidence here suggests that rather than reflecting the ethical and moral dilemmas identified in Chapter Two, it instead reflects a lack of understanding on the part of the professionals involved about the changing expectations families have over time for their child's quality of life, and the different dimensions to the way in which quality of life is understood by families. It can also reflect a lack of awareness on the part of parents, who admit that when their child is first diagnosed with a life-limiting condition it can be hard to conceptualise what sort of quality of life their child might have. Where professionals attend to the features of decision-making for children and young people with life-limiting conditions, and take the time to establish relationships of mutual respect with both parents and children, conflict between professionals and parents is therefore less likely to occur.

In contrast, the more internal conflict that is experienced by parents and young people as they weigh up the potential benefits and drawbacks of life-sustaining technologies is not something that can always be prevented, partly because it is caused by the on-going uncertainty families experience regarding their child's prognosis. This inner conflict is also related to the uncertainty families experience as they make decisions for which the outcomes over the long-term are difficult to predict. Moreover, the feelings of regret and uncertainty families can experience at the time of making a decision can last for many years, and are sometimes re-visited by families as they develop expertise and begin to understand more about their child's quality of life.

Inner conflict can be experienced regardless of the outcomes, and is sometimes caused by the lack of perceived choice in the decision that is made. This can explain why parents are committed to forming their own opinion about a decision, even when they believe the decision has already been made due to the risks involved in not agreeing to a gastrostomy or ventilation. Although the lack of perceived choice is related to the decision under consideration, it can also be influenced by the information and resources made available to families, and to the way a recommendation is perceived, with the strength of recommendations appearing to fall along a continuum with what is viewed as a suggestion at one end and a command at the other.

10.4 Underpinning Decisions by Quality of Life

Quality of life is central to the decisions families make, and enabling children to have the best possible quality of life given the limitations and difficulties they will experience underpins decision-making, a finding supported by the existing literature concerned with how families understand quality of life for children with complex health care needs (Rabiee et al., 2005a, Kirk, 2007). The study identifies a contrast between the views of parents and young people regarding quality of life, with parents emphasising the importance of minimising the pain and distress alongside the aim to ensure that their child is happy and able to take part in the world around them. Young people on the other hand focus on the latter, discussing the use of technological support with regards to the potential limitations on their ability to attend school or college, to spend time with friends, and to engage in social and other activities. Rabiee et al. (2005a) also identify this difference.

By examining how quality of life is assessed for decision-making, this study has been able to draw out the fluidity and complexity of quality of life, and the continual re-adjustments and expectations families make as a child's condition progresses. The study also draws attention to the different elements that make up quality of life when considering decisions that will affect it and provides a new insight into the conflicting nature of the decisions families make for children with progressive and

life-limiting conditions. This was discussed in Chapter Seven, which identifies the difficulties of making a decision that has the potential to enhance one aspect of quality of life, yet at the same time have an adverse effect on another.

Parents and young people can therefore find themselves in conflict about the decision they are asked to make when they have to give up one aspect of quality of life for another, swaying between one position and another as they weigh up the right choice to make. This conflict, while not explicitly identified in the existing literature, is in part reflected by studies that examine the impact of technological support, which draws out the combination of benefits and drawbacks for both children and their parents (Heaton et al., 2003; Kirk and Glendinning, 2004). Although this literature identifies the considerable impact of caring for a technology-dependent child, and the additional and often technical tasks parents take on, the parents in this study did not identify this as a decision factor, instead focusing on the quality of life for their child and the potential impact technological support will have on this.

The existing literature also identifies the potential conflict regarding the uncertainty about the long-term benefits of prolonging the lives of children and young people with progressive conditions (Wright et al., 2009), and the impact over time on their quality of life as their condition deteriorates. Although prognostic uncertainty is identified as a feature in many of the decisions families make, not all the parents and young people included an assessment of the long-term implications, and young people in particular tended to focus on the immediate benefits to their quality of life. This point raises implications about the ability of young people, and in some cases parents to make informed decisions about technological support if they do not include an assessment of the long-term benefits of a proposed intervention given the life-limiting and progressive nature of their condition.

10.5 Shared Decision-Making as a Model for Practice

This research was guided by the conceptual framework of shared decision-making developed by Charles et al. (1999), which was deemed flexible enough to accommodate the triadic nature of decision-making in the paediatric setting, and to capture the complexity surrounding decisions about technological support identified from the literature review. The framework was selected in part because of the trend in policy and practice to recommend such an approach, but also because of the applied objectives for this research to identify the information and support needs of families for decision-making, and to explore the suitability of shared decision-making as a model for exploring how treatment decisions are made for children and young people with life-limiting conditions.

The findings from this study indicate that although the framework reflects certain aspects of treatment decision-making, it does not capture the whole decision process. This is partly because it fails to acknowledge that decisions about technological support, and other decisions families must make for their child are not made in a dyad or triad vacuum, but in an existing health care system that families must interact with when their child is diagnosed with a life-limiting condition. This reveals the central weakness of adopting the model of shared decision-making in practice, because by attending to the exchange of information, the process of deliberation, and the concept of decisional control, the model focuses solely on the physician-patient interaction (Edwards and Elwyn, 2006). By doing so, the role of decision features and process factors identified in this research are excluded.

A further weakness in the model of shared decision-making can be found in its assumption that the recommendation a physician makes equals the point at which the process of decision-making begins. From analysing the retrospective accounts of parents and young people, it has been possible to uncover the wider decision environment within which the actual process of deciding takes place, and this wider process includes within it the series of events and decisions associated with the

symptoms leading up to a recommendation, and in some cases the time between making a decision and the actual initiation of technological support.

Taking into account the findings presented here that not only is there considerable variation in how a recommendation is given to families, but more importantly that the decision process does not, for many families begin at this point, the adoption of a model underpinned by this assumption is not necessarily helpful. Including what comes before a recommendation is essential for decisions about technological support because the background provides important clues about not only the family's experience, views, and knowledge, but in some cases the position they may already have adopted regarding the decision itself.

The model of shared decision-making is however useful in its separation of the different elements of information exchange, deliberation, and decisional control, which are reflected in families' accounts of weighing up the potential impact of technological support on their child's quality of life, and are seen to evolve over time as parents and young people gain confidence and capacity for decision-making. The findings from this research show that while both parents and young people wish to share the process of decision-making with valued professionals, they also emphasise the importance of retaining decisional control, and other research reveals a similar distinction (Edwards and Elwyn, 2006; Knopf et al., 2008). Because the framework of shared decision-making does not assume that patients will be involved to the same extent throughout the different stages of the decision process, it can therefore be used to illustrate the variation in participation.

Although very few decisions reflect the model of shared decision-making when taking into account the wider environment that influences the decision, the actual process of deciding, in other words the process of weighing up the benefits and drawbacks to a proposed intervention, does reflect the model of shared decision-making proposed by Charles et al. (1999), and the guiding principles that underpin it. In fact, the model developed from this research confirms that when families are enabled to make an informed choice, families do engage in a process of shared

decision-making, which is shared between parents and professionals, and over time young people too as their capacity for decision-making develops. In addition, the priority given to expert opinion as a type of information for decision-making also aligns itself with the model of shared decision-making, which views the phase of information exchange as a two-way process between the medical expert and the patient, each bringing knowledge and information about the decision to be made (Charles et al., 1999).

This study revealed variation in the number of professionals involved, and for some children decisions are made between the family and a single trusted professional, who has the relevant expertise and knowledge to enable families to make an informed decision. For other families the involvement of an array of healthcare and other professionals can be confusing, leading to conflict and misunderstanding, and with no single professional that the family is able to develop a relationship with. However not all families share the decision with healthcare professionals and instead, having made a decision, access the system in order that the decision they have made can be followed up. This finding has implications about the growing assumption within the literature concerned with treatment decision-making that 'sharing' the decision with professionals is the best approach (Edwards and Elwyn, 2006). It also raises questions about the recommendation in the literature concerned with life-limited children that in order to improve the process of decision-making for families a multi-disciplinary approach is required (Rouse et al., 2002; McHattie, 2005; Todd et al., 2005).

The model developed from the research allows for this heterogeneity, avoiding assumptions about who should be involved and to what extent, and instead focusing on the wide range of interactions families have with the system, and the key factors about the role of professionals and information that enable rather than constrain families as they make decisions, and therefore enhance the perceived choice in the decisions families make.

10.6 Perceived Choice in Decision-Making

Perceived choice is identified as a key concept in treatment decision-making, and is represented as an outcome of the decision process. The combined influence of decision factors, decision features, and process factors affects the perceived choice families have, and there is a distinction between 'choice' regarding the actual decision, and 'choice' that is related to the family and healthcare system. The study found that for decisions in which the option to initiate technological support clearly outweighs the option to do nothing, the choice of doing nothing is not viewed by families as a real option, and subsequently there is no perceived choice in the decision they have to make. However, this does not mean that there is no decision process, and families still describe the range of decision factors they draw on to make sure the decision that for some families has already been reached due to the lack of choice, is in the best interests of the individual child.

Sometimes though, families do not always perceive the decision they make about technological support as a choice until after the outcomes of the decision are realized. This is partly because in being presented with a single option, it can be difficult for them to recognise they are making a decision that involves two choices, the option to do nothing, and the option to proceed. In fact, the findings show that families do engage in a process of considering two options, firstly assessing whether their child needs technological support, in other words weighing up the option of doing nothing, and secondly considering the potential outcomes of technological support, in other words assessing the option of doing something. However, families do not always view decisions as a choice, and the research identifies 'the recommendation' as a key factor that can influence how families understand the options available. It is essential that professionals are aware of the significance this moment has, and the way in which a recommendation can be interpreted based on how the recommendation is relayed.

Although this research focuses on the factors identified from families' narratives of the decision process, it is important to draw attention to some of the other possible

constraints to making informed choices, and the professional and educational background of parents, and their ability to access and process information for decision-making was identified by families in this research. However, some of the broader inequalities that exist including gender, ethnicity, family income, and religion are also likely to influence the decision process, and this is discussed by Fotaki et al., (2005) who review the literature on patient choice. Housing too is a potential factor, with families of disabled children more likely to live in poor housing with insufficient equipment and adaptations (Cavet, 2009). These factors have the potential to influence both the way in which families make decisions, and the options presented to them by the professionals involved, who may make value judgements about a family's ability to cope with technological support. Further research would be fruitful in exploring how these wider inequalities affect decision-making at the individual level.

10.7 Study Contribution

This study provides new evidence about the decision processes involved in the consideration of technological support for children and young people with life-limiting conditions, drawing attention to the key factors families include as they weigh up the appropriateness of a proposed intervention, and the different and at times conflicting elements of quality of life that underpin the process. The research also contributes new knowledge to the field of decision-making, in particular the stream of work concerned with healthcare decisions, offering a new model of treatment decision-making that, unlike other conceptual models introduced in Chapter Three, has been developed from the analysis of primary data about the lived experience of making complex treatment decisions.

The ecological model developed from the research provides an alternative understanding of the way in which treatment decisions are made by incorporating the wider context and infrastructure that exists around the decision, and offering an explanation of how this interacts with the decision process itself. This approach is in stark contrast to many of the existing decision theories and concepts, which

continue to focus on decision-making at the individual level, for example considering how information is used for decision-making, and exploring the role of cognition and emotion in the decision process (see Beresford and Sloper (2008) for a review of the decision theories on choice and decision-making).

This study draws attention to the combined influence of decision features, decision factors, and process factors on the choice families believe they have in the decisions they make, expanding our understanding of the relationship between choice and decision-making with the concept of 'perceived choice'. The study also contributes new knowledge about the wider constraints to making informed choices, which in the literature tends to be focused around information for decision-making and the way in which information is provided or not provided to individuals as they weigh up a recommended treatment or intervention. Like other research, this study identifies the key role of information for decision-making, however it also provides new evidence about the types and sources of information families would like in order to become informed.

10.8 Study Strengths and Limitations

In considering the scope of the research, this was identified to be the first empirical study to examine decision processes around the use of technological support, and although this was an exploratory piece of research the study was underpinned by robust aims and objectives developed from what is known about decision-making for life-limited children and young people. Furthermore, the quality and credibility of the study was carefully considered throughout the research process, with particular attention to maintaining conceptual and methodological rigour.

However, considering the complexity of decision-making identified from this research, and the use of a single research site from which to draw the sample, it is possible that the study has not been able to capture the full range and diversity of views on the decision processes around the use of technological support. The use of a purposive sample and the efforts made to achieve data saturation go some way to

addressing this, and indeed the data generated is of great depth and richness, and this can be seen in the findings and model derived from the analytical process.

The main limitation of this study relates to the potential imbalance between the views of parents and young people due to the small sample of young people taking part in the research. Consequently, the views of young people need interpreting with care, and although the model has the potential to inform practice, further work is required to make sure that it adequately reflects the experience of children and young people with life-limiting conditions as well as their parents.

10.9 The Need for Further Research

In view of the important contribution to knowledge the findings from this research have to offer and the study's strengths and limitations, further work in this area of decision-making is required if we are to learn more about the ecological nature of decision-making among different patient groups, and for the model of treatment decision-making developed from this study to become a useful learning tool for policy and practice.

A number of research questions have arisen as a result of this work.

- The voices of young people in this work are limited, and more research is needed to further our understanding of the experience of young people in decision processes, and about how they themselves engage with professionals and the healthcare system as decisions are made. The specific concerns relating to the transition to adult services requires further work. What was clear from this study is that the unique family dynamic associated with this patient population, which extends into adulthood due to the increasing dependence of young people with life-limiting conditions, does not sit comfortably inside a health care system underpinned by the autonomous adult patient. Therefore, learning more about the evolving role of parents as well as young people as they make the transition to adult services is identified as an area for future research.

- This research collected data about two forms of technological support, and further work looking at the decision processes around more and less invasive procedures have the potential to enhance our understanding of the decision processes in the use of technological support. Young people referred to the decisions they made regarding spinal rods, due in part to the invasiveness of the procedure, and the introduction of spinal rods at an age when young people are beginning to participate more actively in decision-making, and exploring this decision therefore has the potential to harness the views of young people.
- The model developed is one that reflects the experience of families in decision-making, and further research is required to explore the model through the eyes of professionals if we are to understand more about the interaction of decision features and process factors. While the data collected from professionals will be helpful in developing appropriate methods to do this, the existing data is not sufficient to capture the experiences of the range of professionals identified to be involved in decision-making.

10.10 IMPLICATIONS FOR PRACTICE

Enabling Families to Make Complex Treatment Decisions

Overall, the study found that although there are both common factors and features associated with the decision process, and key factors families use in weighing up the use of technological support, decisions are in fact extremely complex, highly context dependent, and unique for each child and family. The study also shows quality of life, which underpins decisions for children and young people with life-limiting conditions, as a fluid and multifaceted concept, with parents' views regarding their child's quality of life informed by the intrinsic knowledge they acquire about their child over a period of many years.

Professionals must therefore acknowledge the individuality of a family's conception of quality of life, and if possible take the time to listen to families as they talk about their child if they are to align themselves as a partner to parents in decisions about a child's care and treatment. Professionals must also recognise that parents' understanding of the quality of life their child can achieve comes with time, and is particularly difficult when parents are in the early phase of their child's condition, or where there is a great deal of prognostic uncertainty. Tuning into a family's understanding of their child's quality of life can therefore draw attention to the changing expectations parents can have as they gain control as carers and decision-makers for their child, and at times when there is a marked deterioration in their child's condition.

Ultimately, differences in opinion about quality of life will exist, and medical professionals will inevitably have an opinion about the nature and appropriateness of technological support, evident in the literature concerning the use of life-sustaining technologies for children and young people. Although this in some cases can cause conflict in decision-making, this study found that parents and young people also welcome these opinions if they are to make informed decisions, and are not surprised that differences in opinion exist. However, professionals must acknowledge that their own views and beliefs regarding technological support are unlike those held by families, because a family's understanding of quality of life relates to the lived experience of caring for an ill child, and through the continuing adjustments they must make regarding the expectations for their child that shape their understanding of the quality of life their child can hope to achieve.

Essentially, professionals are identified to have a key role in enabling families to make informed choices, which they can achieve by paying attention to the potential impact of process factors, and in recognising the different stages parents can experience in their journey of caring for a child with a life-limiting condition. A family's individual approach to decision-making will influence their experience of the process, and how they interact with professionals in making a decision for their child. The research also sheds light on the parent child dynamics in decision-making,

which will be at different stages and phases for individual families. The point at which parents have reached as decision makers for their child will also influence their ability to understand their child's condition, the needs they might have for information, their view of healthcare professionals, and their ability to take decisional responsibility.

The background to decisions is also important, because when parents arrive at the recommendation having been told over many months or years that they are being paranoid or neurotic, or that their child's symptoms are not as severe as families understand them to be, they may be angry and resistant, and therefore find it difficult to trust what they are now being told. Enabling families to have time, where possible, is therefore essential. This remains the case whether or not the benefits to a proposed intervention clearly outweigh the risks, and as a result there is little perceived choice in the decision a family is asked to make. In these cases, parents and young people still emphasise the importance of engaging in the decision process in a meaningful way, and to do so they need time to acquire information, and to discuss the choices available to them in order to make an informed decision.

Ultimately, being able to access the right information for decision-making is essential if parents and young people feel enabled to make the right choices, regardless of whether the process resembles the model of paternalism or informed choice. Families do not have to be left with unanswered questions, or feelings of uncertainty due to the lack of information. The decisions they make are already surrounded by uncertainty, and the decision factors identified in this study illustrate the efforts parents and young people make to become informed in order to reduce the uncertainty during the decision process.

Given the emphasis among parents on becoming informed and gaining control, it might be easy to make assumptions that parents require information rather than medical opinion. However, one of the key findings from this research is the priority given to this type of information, with parents and young people consistently

identifying medical expertise as the most important type, followed closely by the views of other young people who have experienced the form of technology they are considering. It is therefore essential that professionals recognise the distinction *between information and opinion for decision-making, because where decisions involve uncertainty, and require families to weigh up a range of potential benefits and risks, they will often seek out expert medical opinion and user feedback regarding the decision they must make.*

Finally, it is important to distinguish between the informed position that some families come to have in order to make a decision, and the underlying values and beliefs that may influence this position. The ‘initial gut reaction’ that some parents and young people describe when a gastrostomy or ventilation is proposed will also affect this process, and while families acknowledge the emotional basis of these feelings, the initial response can be felt very strongly by families. Helping young people and parents understand the basis of this feeling is therefore important, because it can endure for many years afterwards and lead parents to continually assess whether or not they have made the right decision, even when the outcomes are positive for their child.

10.11 IMPLICATIONS FOR POLICY

The Model of Informed Choice – An Illusory and Unhelpful Policy Goal?

This study reveals that above all, parents and young people want to make the right decisions. However, in doing so they recognise that sometimes there is little choice in the decisions they have to make. Nevertheless, they still wish to be offered decisional control, and to be sufficiently informed if they are to proceed with an intervention that brings with it both benefits and drawbacks. This finding provides a strong argument for continuing to underpin treatment decision-making by the process of informed consent, because in placing a duty on the healthcare system to ensure families are informed to make decisions, the process enables parents and young people to form their own opinion about the intervention being proposed, regardless of the perceived choice in the decision they are asked to make.

The process of informed consent however is not the same as the concept of informed choice, and it is this model which continues to drive healthcare policy and has permeated practice at the individual level, altering the dynamics between professionals and patients, and leading to assumptions about the preferences of individuals for participating in decision-making, and increasing patients' expectations of choice. The challenges facing healthcare professionals as decision-making continues to be underpinned by the notion of informed choice are therefore significant, particularly in light of the evidence presented here that choices are constrained not only by the decision itself, but by features associated with the patient population and by the system in which professionals work and the role they themselves play in the decision process.

Although informed choice has at its core the process of informed consent, the model is underpinned by the concept of 'patient choice', and drawing on the evidence here it is argued that rather than promoting patient choice as the cornerstone of the NHS, we should instead re-conceptualise and expand our understanding of the concept of informed consent, which is already enshrined in law and embedded in practice. By doing so the focus moves away from participation and decisional control, and moves towards ensuring patients are enabled to make informed decisions about their condition. It also offers a more pragmatic approach to decision-making than what is currently offered through the model of informed choice, because patients are currently led to believe that choices are readily available when in fact this study identifies that this is not a realistic goal.

The model presented in this research is important because it does not make assumptions or value judgements about the best model to adopt when making treatment decisions, or indeed whether to conceptualise the process in such a prescriptive way at all. Furthermore, by identifying the role of systems in the process of decision-making, the model has the potential to bring out the challenges and difficulties experienced by those involved without it being targeted at the individual level. The model also moves beyond the current fixation on participation, which is increasingly seen as the means by which patients are enabled to make

informed choices, and sometimes to the exclusion of all other aspects of decision-making. As this research demonstrates, participation is only one aspect of a wider process, and where professionals take the time to listen to parents and children, and adopt a flexible approach to their role in decision-making, then underpinning decision-making by distinct models is unhelpful, reflecting neither the reality or complexity of treatment decision-making, nor the priorities identified by families in this research.

APPENDICES

Decisions Processes in the Use of Technological Support for Children and Young People with Life-Limiting Conditions

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Appendix A – Information Sheets: Bereaved Parents

Information leaflet – bereaved parents v2 30/09/08

THE UNIVERSITY *of York*



RESEARCH PROJECT

Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions

Information leaflet for parents/guardians

Would you like to help?

We would like to invite you to take part in a research project involving twenty families who have been involved in a decision for their child to have a gastrostomy or to use a mechanical ventilator during the last three years.

We would like to talk to some parents who made this decision some time ago, before their child died, as it is important that their perspectives are included in the study. We understand that your child has died since you made this decision, and you may not want to talk about this. However, if you are willing to think about it please read this leaflet and then let us know whether or not you might be interested in taking part. You may want to discuss the project with someone before you decide.

We are contacting you through [RECRUITING ORGANISATION].

What is the study about?

The project is looking at how decisions about gastrostomy and assisted ventilation for children are made. This includes looking at the information and support parents and children are provided with to help them make a decision, and how they have been helped to make a decision by professionals, for example, nurses or doctors or staff from voluntary organisations.

We would like to talk to you to find out about your experience of making a decision about gastrostomy and / or ventilation for your child, and we would like to explore what were the factors and who were the people that influenced the decision being made. The information you and other families taking part in the study provide will help us find out what information and support families need to help them make these decisions.

What is involved?

We would like to interview you, if we may. The researcher would visit you at home (or another location if you prefer) to carry out the interview. During the interview, we would ask you about your experience of making a decision about a gastrostomy and / or assisted ventilation for your child, and about the information and support you received to assist you and your family. We are particularly interested in how you and your family were involved in the decision, and which professionals you felt supported you at this time.

We expect the interview will last about an hour. In two-parent families, we would like to talk to both parents together or just one parent, as you choose. If you feel that another family member or main carer was really important in the decision about gastrostomy and / or ventilation we would like to talk to them, if they wish. If you agree, we will record the interview. However, if you do not wish to be recorded, we will take notes.

Interview with a professional who has helped with decision making

Health care and other professionals are often involved in helping parents and young people make decisions about their care and treatment, and they may provide information or offer advice. If this is the case with you, we may ask your permission to contact someone who you believe was really important in helping you make the decision about a gastrostomy and / or ventilation for your child. We would like to find out about their role in helping families make these decisions.

Do I have to take part?

No, you do not have to take part.

If you do decide to take part, you will be asked to sign a consent form before the interview. If you later change your mind, you can withdraw at any time. You do not need to give a reason. Whether or not you take part will not affect any services you or your family receive.

Will you tell anyone else what I say?

You have received this invitation via [RECRUITING ORGANISATION], and if you do decide to take part, they will be told. However, they will NOT be told what you say in the interviews. Only members of the research team will know your personal views. No names will be mentioned in the reports of our work and care will be taken so that individuals cannot be identified. All records will be kept locked up and destroyed five years after the study has been completed. Only the researcher and her supervisors will have access to any records from the study.

Although we won't tell others what you've told us, you are of course free to tell others what you said, if you want to.

How will the information I provide be used?

We will examine the information we collect in this study, and write reports on what is found. This will include findings and recommendations about any of the difficulties families experience in making decisions about gastrostomy or assisted ventilation, and the sorts of information and support they need during this process.

On your request, we will send you a summary of the results when the project is finished. A report will be sent to the relevant government departments and organisations involved in the care of children and young people with progressive conditions. We hope that the report will be used by them in planning information and support for families in the future.

You will not be identified in any of the publications.

What are the possible benefits and risks of taking part?

This is a chance for you to tell us about the experience of making a decision about gastrostomy and / or ventilation for your child, and the information and support you as parents needed. You can also tell us who helped you make the decision, and help us identify areas in which the support families receive could be improved. What you tell us is important. It will help us try and improve services for children and young people and their families.

The study only involves talking to a researcher. If you do not want to answer any questions you do not have to. If you become upset, you will be given the option of having a break or stopping the interview. If you require support after an interview, the researcher will help you find appropriate support if needed.

Expenses and payments

You will not be paid for taking part in this study. We will pay for any child-care costs or travel expenses incurred as a result of taking part in the project.

Who is the researcher?

Jo Nicholson is a postgraduate research student based in the Social Policy Research Unit at the University of York. She has relevant training and experience in conducting research and interviews, and will be supervised by Prof Tricia Sloper at the Social Policy Research Unit and Dr Jan Aldridge at Martin House Children's Hospice.

Has the study been reviewed and approved?

This study has been independently reviewed, and it has been approved by an ethics committee [ADD NAME OF MREC AND REF NUMBER].

What do I have to do next?

If you might be interested in taking part, please fill in and return the response slip enclosed. Jo Nicholson will then phone you and tell you more about the project and you can ask any questions about it. If we do not hear from you, you may receive a reminder invitation.

If you decide to take part, we will arrange a time to come and talk to you, and write to confirm this. If you want to change the time or date of the interview for any reason, or change your mind about taking part at any stage, you may do so.

Our contact details

If you would like any further information about the study, please fill in the response slip and Jo Nicholson will contact you. Or you can phone Jo at the Social Policy Research Unit, York University on 01904 321969. You may also like to look at our website at: www.york.ac.uk/inst/spru/

If the reply-paid envelope is missing, please return the slip to:
Jo Nicholson at Social Policy Research Unit, University of York,
Heslington, York, YO10 5DD.

Appendix A – Information Sheets: Parents of Young Adults (age 16 and over)

Information leaflet –parents of young people 16+ v2 30/09/08

THE UNIVERSITY *of York*



RESEARCH PROJECT

Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions

Information leaflet for parents/guardians

Would you like to help?

We would like to invite you to take part in a research project involving twenty families of children with progressive conditions. Please read this leaflet and let us know whether or not you are interested in taking part in the study. You may want to discuss the project with someone before you decide.

What is the study about?

The project is looking at how decisions about gastrostomy and assisted ventilation for children and young people are made. This includes looking at the information and support families are provided with to help them make a decision, and how they have been helped with a decision by professionals, for example, nurses or doctors or staff from voluntary organisations.

We would like to talk to you to find out your experience of making a decision about gastrostomy and / or ventilation for your child, and we would like to explore what are the factors and who are the people that influence the decisions being made. The information you and other families taking part in the study provide will help us find out what information and support families need to help them make decisions about gastrostomy and / or ventilation.

Why have I been invited to take part?

We are contacting you through [RECRUITING ORGANISATION], as your son or daughter is aged between seven and twenty-four, and you have made a decision about a gastrostomy and / or assisted ventilation for your child in the last three years. Your son or daughter has also been invited to take part in the study.

What is involved?

We would like to interview you, if we may. The researcher would visit you at home (or another location if you prefer) to carry out the interview. During the interview, we would like to ask you about your experience of making a decision about a gastrostomy and / or assisted ventilation for your child, and about the information and support you received to assist you and your family. We are particularly interested in how you and your family were involved in the decision, and which professionals you felt supported you at this time.

We expect the interview will last about an hour. In two-parent families, we would like to talk to both parents together or just one parent, as you choose. If you feel that another family member or main carer was really involved in the decision about gastrostomy and / or ventilation, we would like to talk to them, if they wish. If you agree, we will record the interview. However, if you do not wish to be recorded, we will take notes.

Interview with children and young people

We would also like to talk to your son or daughter about the same issues as yourself, either directly after your interview, or on a later date (you and your child can choose). We have sent some information for your child, so they can decide if they would like to take part or not.

Interview with a professional who has helped with decision making

Health care and other professionals are often involved in helping parents and young people make decisions about their care and treatment, and they may provide information or offer advice. If this is the case with you, we may ask your permission to contact someone who you believe was really important in helping you make the decision about a gastrostomy and / or assisted ventilation. We would like to find out about their role in helping families make these types of decisions.

Do I have to take part?

No, you do not have to take part. *If you decide not to that's OK.* If you do decide to take part, you will be asked to sign a consent form before the interview. If you later change your mind, you can withdraw at any time. You do not need to give a reason. Whether or not you take part will not affect any services you or your family receive.

Will you tell anyone else what I say?

You have received this invitation via [RECRUITING ORGANISATION], and if you do decide to take part, they will be told you have agreed to take part. However, they will NOT be told what you say in the interviews. Only members of the research team will

Information leaflet –parents of young people 16+ v2 30/09/08

know your personal views. Your name will NOT be mentioned to others or published in the reports of our work. If we use any quotes from what people have told us, no names will be mentioned. All records will be kept locked up and destroyed five years after the study has been completed.

Although we won't tell others what you've told us, you are of course free to tell others what you said, if you want to.

How will the information I provide be used?

We will examine the information we collect in this study, and write reports on what is found. This will include findings and recommendations about some of the difficulties families experience in making decisions about gastrostomy and assisted ventilation, and the sorts of information and support they need during this process.

On your request, we will send you a summary of the results when the project is finished. A full report will be sent to the relevant government departments and statutory /voluntary organisations. We hope that the report will be used by them in planning information and support for families in the future.

You will not be identified in any of the publications.

What are the possible benefits and risks of taking part?

This is a chance for you to tell us about the decision you and your family made, and the information and support you as parents need to help with such decisions. You can also tell us who helped you make the decision, and help us identify areas in which the support families receive could be improved. What you tell us is important. It will help us try and improve services for children and young people and their families.

The study only involves talking to a researcher. If you do not want to answer any questions you do not have to. If you become upset, you will be given the option of having a break or stopping the interview. If you require support after an interview, you can contact the researcher who will help you find appropriate support if needed.

Expenses and payments

You will not be paid for taking part in this study. We will pay for any child-care costs or travel expenses incurred as a result of taking part in the project.

Who is the researcher?

Jo Nicholson is a postgraduate research student based in the Social Policy Research Unit at the University of York. She has relevant training and experience in conducting research and interviews, and will be supervised by Prof Tricia Sloper at the Social Policy Research Unit and Dr Jan Aldridge at Martin House Children's Hospice.

Has the study been reviewed and approved?

This study has been independently reviewed, and it has been approved by an ethics committee [ADD NAME OF MREC AND REF NUMBER].

What do I have to do next?

If you are interested in taking part, please fill in and return the response slip enclosed. Jo Nicholson will then phone you and tell you more about the project and you can ask any questions about it. If we do not hear from you, you may receive a reminder invitation.

If you decide to take part, we will arrange a time to come and talk to you, and write to confirm this. If you want to change the time or date of the interview for any reason, or change your mind about taking part at any stage, you may do so.

Our contact details

If you would like any further information about the study, please fill in the response slip and Jo Nicholson will contact you. Or you can phone Jo at the Social Policy Research Unit, York University on 01904 321969. You may also like to look at our website at: www.york.ac.uk/inst/spru/

If the reply-paid envelope is missing, please return the slip to:
Jo Nicholson at Social Policy Research Unit, University of York,
Heslington, York, YO10 5DD.

Appendix A – Information Sheets: Parents of Children under the Age of 16

Information leaflet – parents of children under 16 v2 30/09/08

THE UNIVERSITY *of York*



RESEARCH PROJECT

Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions

Information leaflet for parents/guardians

Would you like to help?

We would like to invite you to take part in a research project involving twenty families of children with progressive conditions. Please read this leaflet and let us know whether or not you are interested in taking part in the study. You may want to discuss the project with someone before you decide.

What is the study about?

The project is looking at how decisions about gastrostomy and assisted ventilation for children are made. This includes looking at the information and support families are provided with to help them make a decision, and how they have been helped with a decision by professionals, for example, nurses or doctors or staff from voluntary organisations.

We would like to talk to you, and to your child if we may, to find out your experience of making a decision about gastrostomy and / or ventilation for your child. We would also like to explore what are the factors and who are the people that influence the decisions being made. The information you and other families taking part in the study provide will help us find out what information and support families need to help them make decisions about gastrostomy and / or ventilation.

Why have I been invited to take part?

We are contacting you through [RECRUITING ORGANISATION], as your son or daughter is aged between seven and twenty-four, and you have made a decision about a gastrostomy and / or assisted ventilation for your child in the last three years.

What is involved?

We would like to interview you and your child, if we may. If your son or daughter does not wish to or can't take part we would still like to invite you to take part if you wish.

and if your child is happy for you to do so. Similarly, if your son or daughter wants to take part but you don't, he or she can take part but you don't have to.

Interviews with parents

The researcher would visit you at home (or another location if you prefer) to carry out the interview. During the interview, we would like to ask you about your experience of making a decision about a gastrostomy and / or assisted ventilation for your child, and about the information and support you received to assist you and your family. We are particularly interested in how you and your family were involved in the decision, and which professionals you felt supported you at this time.

We expect the interview will last about an hour. In two-parent families, we would like to talk to both parents together or just one parent, as you choose. If you feel that another family member or main carer was really involved in the decision about gastrostomy and / or ventilation, we would like to talk to them, if they wish. If you agree, we will record the interview. However, if you do not wish to be recorded, we will take notes.

Interviews with children and young people

We would also like to talk to your child about the same decision, either directly after your interview, or on a later date (you and your child can choose). The interview with young people would be shorter. We will adapt the interview to suit the young person's abilities and use pictures and symbols as well as words.

We have enclosed some information for you to give to your child if you are happy to do this, so they can decide if they would like to take part. As your child is under the age of 16 we will need parental consent for him/her to do so.

Interview with a professional who has helped with decision making

Health care and other professionals are often involved in helping parents and young people make decisions about their care and treatment, and they may provide information or offer advice. If this is the case with you, we may ask your permission to contact someone who you believe was really important in helping you make the decision about a gastrostomy and / or assisted ventilation. We would like to find out about their role in helping families make these types of decisions.

Similarly, we would like to ask your son/daughter if he/she has been helped by someone and if we could also interview them.

Do I have to take part?

No, you do not have to take part. *If you decide not to that's OK.* If you do decide to take part, you will be asked to sign a consent form before the interview. If you later change your mind, you can withdraw at any time. You do not need to give a reason. Whether or not you take part will not affect any services you or your family receive.

Will you tell anyone else what I say?

You have received this invitation via [RECRUITING ORGANISATION], and if you do decide to take part, they will be told you have agreed to take part. However, they will NOT be told what you or your child says in the interviews. Only members of the research team will know your personal views. Your name will NOT be mentioned to others or published in the reports of our work. If we use any quotes from what people have told us, no names will be mentioned. All records will be kept locked up and destroyed five years after the study has been completed.

Although we won't tell others what you've told us, you are of course free to tell others what you said, if you want to.

How will the information I provide be used?

We will examine the information we collect in this study, and write reports on what is found. This will include findings and recommendations about some of the difficulties families experience in making decisions about gastrostomy and assisted ventilation, and the sorts of information and support they need during this process.

On your request, we will send you a summary of the results when the project is finished. A full report will be sent to the relevant government departments and statutory /voluntary organisations. We hope that the report will be used by them in planning information and support for families in the future.

You will not be identified in any of the publications.

What are the possible benefits and risks of taking part?

This is a chance for you to tell us about the decision you and your family made, and the information and support you as parents need to help with such decisions. You can also tell us who helped you make the decision, and help us identify areas in which the support families receive could be improved. What you tell us is important. It will help us try and improve services for children and young people and their families.

The study only involves talking to a researcher. If you do not want to answer any questions you do not have to. If you become upset, you will be given the option of

having a break or stopping the interview. If you require support after an interview, you can contact the researcher who will help you find appropriate support if needed.

Expenses and payments

You will not be paid for taking part in this study. We will pay for any child-care costs or travel expenses incurred as a result of taking part in the project.

Who is the researcher?

Jo Nicholson is a postgraduate research student based in the Social Policy Research Unit at the University of York. She has relevant training and experience in conducting research and interviews, and will be supervised by Prof Tricia Sloper at the Social Policy Research Unit and Dr Jan Aldridge at Martin House Children's Hospice.

Has the study been reviewed and approved?

This study has been independently reviewed, and it has been approved by an ethics committee [ADD NAME OF MREC AND REF NUMBER].

What do I have to do next?

If you are interested in taking part, please fill in and return the response slip enclosed. Jo Nicholson will then phone you and tell you more about the project and you can ask any questions about it. If we do not hear from you, you may receive a reminder invitation.

If you decide to take part, we will arrange a time to come and talk to you, and write to confirm this. If you want to change the time or date of the interview for any reason, or change your mind about taking part at any stage, you may do so.

Our contact details

If you would like any further information about the study, please fill in the response slip and Jo Nicholson will contact you. Or you can phone Jo at the Social Policy Research Unit, York University on 01904 321969. You may also like to look at our website at: www.york.ac.uk/inst/spru/

If the reply-paid envelope is missing, please return the slip to:
Jo Nicholson at Social Policy Research Unit, University of York,
Heslington, York, YO10 5DD.

Appendix A – Information Sheets: Young Adults (age 16 and over)

Information leaflet – young people (full text) v2 30/09/08

THE UNIVERSITY *of York*



Research Project **Decisions about gastrostomy or assisted ventilation** **for children and young people**

Information leaflet for young people

Would you like to help?

We would like to invite you to take part in a research project involving twenty families of young people who have made a decision about having a gastrostomy or using a ventilator. Please read this leaflet and let us know whether or not you are interested in taking part in the study. You may want to discuss the project with someone before you decide.

What is the study about?

The project is looking at how decisions about gastrostomy and assisted ventilation are made. This includes looking at the information and support families and young people are provided with to help them make a decision, and how they have been helped with a decision by key professionals, for example nurses or doctors or staff from voluntary organisations.

We would like to talk to you, and to your parents if we may, to listen to your views about the decision for you to have a gastrostomy or to use a mechanical ventilator. We would also like to find out what information and support you received, and how other people may have helped you.

The information you and other people taking part in the study provide will help us find out what information and support families and young people need to help them make decisions about gastrostomy and / or assisted ventilation.

Why have I been invited to take part?

We are contacting you through [RECRUITING ORGANISATION], as you are aged between 7 and 24, and you have been involved in making a decision about having a gastrostomy or using a ventilator.

What is involved?

We would like to interview you and your parents if we may. You will be interviewed separately to your parents. If your parents do not wish to take part, we would still like to invite you to take part, but if you are aged 15 or less you will need your parent's permission. Similarly, if your parents want to take part but you don't, they can but you don't have to.

Interviews with young people

We would like to visit and talk with you at home, or somewhere else if you prefer. If you would like to see us, you can choose to talk to us on your own or with someone else present.

During the interview, we would like to ask you about getting a gastrostomy or using a ventilator. We are particularly interested in what information you were given, and how other people helped you learn more about using a ventilator or having a gastrostomy.

We expect the interview will last about an hour. If you agree, we will record the interview. However, if you don't want to be recorded, we will take notes.

Interviews with parents

We would also like to talk to your parents about the same issues as yourself, either directly before your interview, or on an earlier date (you and your parents can choose). We have sent some information for your parents, so they can decide if they would like to take part or not.

Interview with another person who has helped with decision making

Sometimes other people - like nurses or doctors - are very involved in helping people to make decisions; for example, they may provide information or offer advice. If this is the case with you, we may ask your permission to contact someone who has helped you and your family learn more about having a gastrostomy or using a ventilator. We would like to find out about their role in helping families make these decisions. If you are aged 15 or less, we would need your parent's permission too.

Do I have to take part?

No, you do not have to take part. *If you decide not to that's OK.* If you do decide to take part, you will be asked to sign a consent form before the interview. You can change your mind at any time without giving a reason. Whether or not you take part will not affect any services or help you or your family receive. If you are aged 15 or less, you will need your parents to sign the consent form too.

Will you tell anyone else what I say?

You have received this invitation via [RECRUITING ORGANISATION], and if you do decide to take part, they will be told. However, they will NOT be told what you say in the interviews. You can choose what you tell us and we will not tell anyone what you have said to us. Your name will NOT be mentioned to others or in the reports of our work. All records will be kept locked up and destroyed five years after the study has finished.

Although we won't tell others what you've told us, you can tell others what you said, if you want to.

How will the information I provide be used?

We will write a report on what we found in this study. This will include some of the difficulties families experience in making decisions about gastrostomy and assisted ventilation, and the sorts of information and support they need.

If you agree, we will send you a summary of the results when the project is finished. Reports on the study will also be sent to organisations that support young people and their families, and we hope it will be used by them in planning information and support in the future.

You will not be identified in any of the publications.

What are the possible benefits and risks of taking part?

This is a chance for you to tell us how you feel about having a gastrostomy or using a ventilator, and the sorts of information and support you and your family need. What you tell us is important. It will help us try and improve services for children and young people and their families.

The study only involves talking to a researcher. If you do not want to answer any questions you do not have to. If you become upset, you will be given the option of

having a break or stopping the interview. If you want support after an interview, the researcher will help you find someone who can help. If you are under 16, we will need your parents' permission to contact anyone for you.

Expenses and payments

You will not be paid for taking part in this study, but we will pay for any travel expenses if you have to travel to take part in the project.

Who is the researcher?

Jo Nicholson is a research student based in the Social Policy Research Unit at the University of York. She has relevant training and experience in interviewing people for research, and will be supervised by Prof Tricia Sloper at the Social Policy Research Unit and Dr Jan Aldridge at Martin House Children's Hospice.

Has the study been reviewed and approved?

This study has been independently reviewed, and it has been approved by [ADD NAME OF MREC AND REF NUMBER].

What do I have to do next?

If you are interested in taking part, please fill in and return the response slip enclosed. Jo will then phone you and tell you more about the project and answer any questions you have. You can then decide whether you would like to take part. If we do not hear from you, you may receive a reminder invitation.

If you decide to take part, we will arrange a time to visit you, and write to confirm this. If you want to change the time or date of the interview, or change your mind about taking part at any stage, that's OK, just let us know.

My contact details

If you would like any more information about the study you can phone Jo at the Social Policy Research Unit, York University on 01904 321969. You may also like to look at our website: www.york.ac.uk/inst/spru/

If the reply-paid envelope is missing, please return the slip to:
Jo Nicholson at Social Policy Research Unit, University of York,
Heslington, York, YO10 5DD.

Appendix A – Information Sheets: Young People Age 12 to 16

Information leaflet – young people (short text) v2 30/09/08

THE UNIVERSITY *of York*



Research project about gastrostomy or assisted ventilation for children and young people

Information leaflet for young people

Can you help me?

I would like to invite you to take part in a research project.

What is it all about?

My name is Jo and I work at the University of York. I talk to children and young people and their families to find out more about the things that matter to them. I would like to listen to your views about getting a gastrostomy or using a ventilator. I would like you to tell me who helps you learn more about a gastrostomy or ventilator, and the information you find helpful.

I want to write a report, which will help the services for young people and families learn more about what they can do to help and support young people like you. I want to improve services for young people and parents but I need you to help me if you can.

This is a chance for you to tell me what matters to you.

What you tell me is important.

What would you have to do if you take part?

I would like to talk with you about getting a gastrostomy or using a ventilator. I can visit you at home, or you may prefer us to meet in a place where there are other people you know and where you can talk in private. If you are interested in taking part in the project, I will contact you again and, if you agree, I will arrange to come and talk with you. I will ask you what support you need to take part and I will try to provide it for you. If

Information leaflet – young people (short text) v2 30/09/08

you would like to see me, you can choose to talk to me on your own, or with someone else present. This could be your mum, dad or guardian, your brother or sister, or a teacher, carer or a friend – you can choose. If you agree, I will record the interview. However, if you don't want to be recorded, I will take notes.

Sometimes, other people – like doctors or nurses – are very involved in helping people to make decisions. I may ask you if I can contact someone who has helped you learn more about a gastrostomy or a ventilator.

It's private and confidential

You can choose what you want to tell me and I will not tell anyone what you have said to me. I will not mention your name in my report.

Do you want to take part?

I hope you will want to talk to me, but this is your choice. I will only come and see you if you want me to. It is OK to say no if this is what you want. If you do decide to take part and then change your mind for any reason, you do not have to take part, and you do not have to tell me why you have changed your mind.

What to do now?

If you think you would like to take part in this project, please can you and your parent or guardian fill in the form attached to this letter, and send it back to me. I will then phone you to answer any questions you want to ask me and, if you agree, we can arrange to meet.

Any questions?

If you would like to talk to me about the project, or if you have any questions about it, please phone, email or write to me.

Jo Nicholson
Social Policy Research Unit
University of York
York YO10 5DD

Telephone: 01904 321969

Email: jn123@york.ac.uk

Appendix A – Information Sheets: Children Age 7 to 11

Information leaflet – young people (simple text) v2 30/09/08

THE UNIVERSITY *of York*



Research project about having a feeding tube or using a ventilator

Information leaflet for young people

Can you help?

Hello, my name is Jo and I am from the University of York.

I am doing a project about children and young people who may need a feeding tube, or who may need to use a machine called a ventilator to help them breathe.

I would like to talk and listen to children and young people about getting a feeding tube.

I would also like to talk and listen to children and young people about using a machine to help them breathe.

I would like to find out about any information you used to learn about a feeding tube or ventilator, and who gave you that information.

I would also like to find out if anyone talked to you about getting a feeding tube or using a ventilator, or helped you learn about it.

The things you and other people tell me will be used to write a report.

The report will not have your name in, and the people who read it will not know who you are.

Would you like to talk to me about this?

I can come to your house or if you like, you can choose another place for us to meet.

If you would like to meet me first, and then decide if you want to talk to me, that is OK.

You can say yes or no. You choose.

If you have a question, please phone or write to me.

Thank you, Jo

Appendix B – Response Forms: Bereaved Parents / Parents of Young Adults

Response form – parents v2 30/09/08

THE UNIVERSITY *of York*

 Social Policy
Research Unit

RESEARCH PROJECT
Decisions about gastrostomy or assisted ventilation
for children and young people with progressive conditions

Parent/guardian's response form

For parent/guardian to complete

	YES	NO
I might be interested in taking part in the project and I am happy for a researcher to contact me.	<input type="checkbox"/>	<input type="checkbox"/>

If you answered 'Yes' above, please complete the rest of the form and return it using the pre-paid envelope to Jo Nicholson. Jo will then telephone you to discuss your participation and answer any questions you may have, before you decide whether or not to arrange to talk to a researcher.

If you answered 'No' and do NOT wish to receive a reminder letter, please fill in your name and address, and return it to Jo Nicholson. Otherwise, you may be sent a reminder letter.

YOUR CONTACT DETAILS. (Please write clearly, in BLOCK letters)

Young person's name: _____

Date of birth: _____

Parent's/guardian's name: _____

Address: _____

Postcode: _____ Telephone: _____

Best time of day to call: _____

E-mail (if applicable): _____

Social Policy Research Unit • University of York • Heslington • York • YO10 5DD
Telephone (01904) 321950 • Text Telephone (01904) 321951

Appendix B – Response Forms: Parents of Children Under 16

Response form – parents and young people under 16 v2 30/09/08

THE UNIVERSITY of York



RESEARCH PROJECT
Decisions about gastrostomy or assisted ventilation
for children and young people with progressive conditions

Parent/guardian's and young person's response form

For parent/guardian to complete

I might be interested in taking part in the project and I am happy for a researcher to contact me.

YES

NO

My child might be interested in taking part and I am happy for a researcher to contact him/her.

For children and young people

I might be interested in taking part in the project and I am happy for a researcher to contact me.

I am happy for my parent / guardian to take part in the project.

If either a parent/guardian or young person answered 'Yes' above, please complete the rest of the form and return it using the pre-paid envelope to Jo Nicholson. Jo will then telephone you to discuss your participation and answer any questions you may have, before you decide whether or not to arrange to talk to a researcher.

If you both answered 'No' and do NOT wish to receive a reminder letter, please fill in your name and address, and return it to Jo Nicholson. Otherwise, you may be sent a reminder letter.

YOUR CONTACT DETAILS. (Please write clearly, in BLOCK letters)

Young person's name: _____

Date of birth: _____

Please turn over...

Response form – parents and young people under 16 v2 30/09/08

Parent's guardian's name: _____

Address: _____

Postcode: _____ Telephone: _____

Best time of day to call: _____

Email (if applicable): _____

Social Policy Research Unit • University of York • Heslington • York • YO10 5DD
Telephone (01904) 321950 • Text Telephone (01904) 321951

Appendix B – Response Forms: Young Adults

Response form – young adults v2 30/09/08

THE UNIVERSITY *of York*



RESEARCH PROJECT Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions

Young adult's response form

For young adult to complete

YES

NO

I might be interested in taking part in the project and I am happy for a researcher to contact me.

If you answered 'Yes' above, please complete the rest of the form and return it using the pre-paid envelope to Jo Nicholson. Jo will then telephone you to discuss your participation and answer any questions you may have, before you decide whether or not to arrange to talk to a researcher.

If you answered 'No' and do NOT wish to receive a reminder letter, please fill in your name and address, and return it to Jo Nicholson. Otherwise, you may be sent a reminder letter.

YOUR CONTACT DETAILS (Please write clearly, in BLOCK letters)

Name: _____

Date of birth: _____

Address: _____

Postcode: _____ Telephone: _____

Best time of day to call: _____

Email (if applicable): _____

Social Policy Research Unit • University of York • Heslington • York • YO10 5DD
Telephone (01904) 321950 • Text Telephone (01904) 321951

Appendix C – Invitation Letters: Bereaved Parents

Invitation letter – bereaved parents v2 30/09/08

HOSPICE HEADED PAPER

LETTER OF INVITATION TO PARENTS/ GUARDIANS RE. TAKING PART IN THE STUDY

Date

Dear [PARENT GUARDIAN]

**Re: Research Project – Decisions about gastrostomy or assisted ventilation
for children and young people with progressive conditions**

I am writing to tell you about a research project that you may be interested in taking part in.

The project is called 'Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions'. It is looking at how decisions about gastrostomy or assisted ventilation are made, and the information and support that is provided to families to help with such decisions. It is hoped that around twenty families will take part in the study, and will include bereaved families and families whose child may be able to take part in the project as well as their parents.

The researcher would like to talk to you to find out your views on the decision you made about gastrostomy and/or assisted ventilation for your child in a single interview expected to last around one hour. If you feel that another member of your family was involved in the decision, and they would like to take part in the project, the researcher would also like to talk to them.

The project is being carried out by Jo Nicholson, a postgraduate research student from the University of York. The project is being supervised by Tricia Sloper from the University of York and Jan Aldridge from Martin House Children's Hospice.

The enclosed Information Leaflet tells you more about the project and what taking part in it involves. Please read the leaflet carefully and let Jo Nicholson know whether or not you are interested in taking part by filling in the enclosed form.

If you have any questions about the project and what taking part involves, please contact either myself on the telephone number above, or you can phone Jo Nicholson at the Social Policy Research Unit (SPRU) on 01904 321969.

Yours sincerely

[NAME/ORGANISATION]

Enc. Information Leaflet for Parents/Guardians
Response form
Pre-paid envelope (1st class) to Jo Nicholson, SPRU

Appendix C – Invitation Letters: Parents of Young Adults

Invitation letter – parents of young people 16+ v2 30/09/08

HOSPICE HEADED PAPER

**LETTER OF INVITATION TO PARENTS/
GUARDIANS RE. TAKING PART IN THE
STUDY**

Date

Dear [PARENT/GUARDIAN]

**Re: Research Project – Decisions about gastrostomy or assisted ventilation
for children and young people with progressive conditions**

I am writing to tell you about a research project that you may be interested in taking part in.

The project is called 'Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions'. It is looking at how decisions about gastrostomy and assisted ventilation are made, and the information and support that is provided to families to help with such decisions.

The researcher would like to talk to you to find out your views on the decision you have made about gastrostomy and/or ventilation for your child in a single interview expected to last around one hour. If you feel that another member of your family was involved in this decision, and they would like to be involved in the project, the researcher would also like to talk to them. Your son or daughter has also been sent an invitation to take part in the project.

The project is being carried out by Jo Nicholson, a postgraduate research student from the University of York. The project is being supervised by Tricia Sloper from the University of York and Jan Aldridge from Martin House Children's Hospice.

The enclosed Information Leaflet tells you more about the project and what taking part in it involves. Please read the leaflet carefully and let Jo Nicholson know whether or not you are interested in taking part by filling in the enclosed form.

If you have any questions about the project and what taking part involves, please contact either myself on the telephone number above, or you can phone Jo Nicholson at the Social Policy Research Unit (SPRU) on 01904 321969.

Yours sincerely

[NAME/ORGANISATION]

Enc. Information Leaflet for Parents/Guardians
Response form
Pre-paid envelope (1st class) to Jo Nicholson, SPRU

Appendix C – Invitation Letters: Parents of Children under the Age of 16

Invitation letter – parents of children under 16 v2 30/09/08

HOSPICE HEADED PAPER

LETTER OF INVITATION TO PARENTS/ GUARDIANS RE. TAKING PART IN THE STUDY

Date

Dear [PARENT/GUARDIAN]

**Re: Research Project – Decisions about gastrostomy or assisted ventilation
for children and young people with progressive conditions**

I am writing to tell you about a research project that you may be interested in taking part in.

The project is called 'Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions'. It is looking at how decisions about gastrostomy and assisted ventilation are made, and the information and support that is provided to families to help with such decisions.

The researcher would like to talk to you and your child, if she may, to find out your views on the decision you have made in a single interview expected to last around one hour. If you feel that another member of your family has been involved in the decision about gastrostomy and/or ventilation, and they would like to be involved in the project, the researcher would also like to talk to them.

The project is being carried out by Jo Nicholson, a postgraduate research student from the University of York. The project is being supervised by Tricia Sloper from the University of York and Jan Aldridge from Martin House Children's Hospice.

The enclosed Information Leaflet tells you more about the project and what taking part in it involves for you and your child. Please read the leaflet carefully. An Information Leaflet has also been enclosed for you to give to your child, if you are happy to do so. Please discuss whether or not you each want to take part. As your child is under the age of 16, she/he will need your permission to take part.

Please let Jo Nicholson know whether or not you and/or your child are interested in taking part by filling in the enclosed form. If you have any questions about the project and what taking part involves, please contact either myself on the telephone number above, or you can phone Jo Nicholson at the Social Policy Research Unit (SPRU) on 01904 321969.

Yours sincerely

[NAME/ORGANISATION]

Enc. Information Leaflet for Parents/Guardians
Information Leaflet for Young People
Response form
Pre-paid envelope (1st class) to Jo Nicholson, SPRU

Appendix C – Invitation Letters: Young Adults

Invitation letter - young adults v2 30/09/08

HOSPICE HEADED PAPER

**LETTER OF INVITATION TO YOUNG PEOPLE
RE. TAKING PART IN THE STUDY**

Date

Dear [YOUNG PERSON]

**Re: Research Project - Decisions about gastrostomy or assisted ventilation
for children and young people with progressive conditions**

I am writing to tell you about a research project that you may be interested in taking part in.

The project is called 'Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions'. It is looking at how decisions about gastrostomy and assisted ventilation are made, and the sort of information and support that is provided to families to help with such decisions.

The researcher would like to talk to you and your parent/guardian if they agree, to find out your views on the decision you and your family have made. Your parent/guardian has also been sent an invitation to take part in the project. You would be interviewed separately to your parents, and an interview is expected to last no more than one hour.

The project is being carried out by Jo Nicholson, a postgraduate research student from the University of York. The project is being supervised by Inca Slogar from the University of York and Jan Aldridge from Martin House Children's Hospice.

The enclosed Information Leaflet tells you more about the project and what taking part involves. Please read the leaflet carefully and discuss it with your parent/guardian if you wish.

Please let the researcher know whether or not you are interested in taking part by filling in the enclosed form.

If you have any questions about the project and what taking part involves, please contact me on the telephone number above, or you can phone Jo Nicholson at the Social Policy Research Unit (SPRU) on 01904 321969.

Yours sincerely

[NAME ORGANISATION]

Enc. Information Leaflet for Young Adults

Response form

Pre-paid envelope (1" class) to Jo Nicholson, SPRU.

Appendix D – Consent Form: Parents

THE UNIVERSITY <i>of York</i>		
<p><i>RESEARCH PROJECT</i> Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions</p>		
<p><i>RESEARCHER</i> Jo Nicholson</p>		
<p>Consent form for parents/guardians</p>		
<p>This form is for you to state whether or not you agree to take part in the study. Please read and answer every question. If there is anything you do not understand, or if you want more information, please ask the researcher.</p>		
	YES	NO
Have you read and understood the information leaflet about the study?	<input type="checkbox"/>	<input type="checkbox"/>
Have you had an opportunity to ask questions about the study?	<input type="checkbox"/>	<input type="checkbox"/>
Do you understand that the information you provide will be held in confidence by the research team?	<input type="checkbox"/>	<input type="checkbox"/>
Do you understand that the information you provide will be used for reports and publications, and that your name will not be identified?	<input type="checkbox"/>	<input type="checkbox"/>
Do you understand that the information you provide will be securely stored by the researcher, and will be destroyed when no longer required?	<input type="checkbox"/>	<input type="checkbox"/>
Do you understand that you may withdraw from the study at any time and for any reason, without affecting any services you receive?	<input type="checkbox"/>	<input type="checkbox"/>
Do you understand that your interview transcript may be requested by a court in the event of any legal action you decide to take relating to your child's care or treatment?	<input type="checkbox"/>	<input type="checkbox"/>
<p>Original copy to be retained by the research team. Copy to be sent to participant by post.</p>		

Do you agree to take part in the study?

If yes, do you agree to your interviews being recorded?

(You may take part in the study without agreeing to this)

All data is held by SPRU in accordance with the Data Protection Act.

Your name (in BLOCK letters): _____

Your signature: _____

Interviewer's name: _____

Date: _____

Original copy to be retained by the research team. Copy to be sent to participant by post.

Appendix D – Consent Form: Young Adults (age 16 and over)

THE UNIVERSITY *of York*



RESEARCH PROJECT

Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions

RESEARCHER – Jo Nicholson

Consent form for young people

Please read and answer every question.

Have you read, or has someone read to you, the 'Information Leaflet for Young People' about the project?

Yes No

Do you understand what the project is about and what taking part involves?

Yes No

Do you understand that the project is strictly private?

Yes No

Do you understand that the information will be used for reports and your name will not be identified?

Yes No

Do you understand that the information will be securely stored and then destroyed when no longer required?

Yes No

Do you know that you can leave the project at any time without giving a reason?

Yes No

Do you understand that a copy of your interview may be requested by a court should any legal action be taken about your care or treatment?

Yes No

Please turn over

Original copy to be retained by the research team. Copy to be sent to participant by post.

Would you like to take part in the project?

Yes No

If yes – is it OK to record your interview?

Yes No

Please write your name here: _____

Please sign your name here: _____

Interviewer's name: _____

Date: _____

Original copy to be retained by the research team. Copy to be sent to participant by post.

Appendix D – Consent Form: Young People age 12 to 16

THE UNIVERSITY *of York*



RESEARCH PROJECT

Decisions about gastrostomy or assisted ventilation for children and young people with progressive conditions

RESEARCHER – Jo Nicholson

Consent form for young people

Please read and answer every question.

Have you read, or has someone read to you, the information leaflet about the project?

Yes No

Do you understand what the project is about and what taking part involves?

Yes No

Do you understand that the project is strictly private?

Yes No

Do you understand that the information will be used for reports and your name will not be identified?

Yes No

Do you understand that the information will be securely stored and then destroyed when no longer required?

Yes No

Do you know that you can leave the project at any time without giving a reason?

Yes No

Please turn over

Original copy to be retained by the research team. Copy to be sent to participant by post.

Would you like to take part in the project? Yes No

If yes - is it OK to record your interview? Yes No

Please write your name here:

Please sign your name here:

As you are aged 15 or less, a parent/guardian also needs to give consent:

Parent/guardian's name (in BLOCK letters):

Parent/guardian's signature: _____

Interviewer's name: _____

Date: _____

Original copy to be retained by the research team. Copy to be sent to participant by post.

Appendix D – Consent Form: Children age 7 to 11

THE UNIVERSITY *of York*



*Research project about having a feeding tube
or using a ventilator*

Researcher – Jo Nicholson

Consent form for children to take part in the project

Please read and answer every question.

Have you read, or has someone read to you,
the 'Information Leaflet' about the project? Yes No

Do you understand what the project is about? Yes No

Do you understand what taking part involves? Yes No

Do you understand that the project is private? Yes No

Do you understand that the information
will be used to write a report and your
name will not be used? Yes No

Do you understand that the information
will be kept safe and then destroyed
when no longer needed? Yes No

Please turn over

Original copy to be retained by the research team. Copy to be sent to participant by post.

Do you know that you can leave the project
at any time without giving a reason?

Yes No

Would you like to take part in the project?

Yes No

If yes - can I record our conversations?

Yes No

Please write your name here:

As you are aged 16 or less, a parent/guardian also needs to give consent:

Parent/guardian's name (in BLOCK letters)

Parent/guardian's signature: _____

Interviewer's name: _____

Date: _____

Original copy to be retained by the research team. Copy to be sent to participant by post.

Appendix E – Interview Topic Guides: Parents

DECISION CONTEXT

- Collect information about the child and family – details of their illness, primary caregivers, family composition
- Collect information about the child’s quality of life - likes and dislikes, things that are important to them, people important in their lives, whether they go to school
- Collect information about the decision to have g-tube / ventilation – where and when the decision was made, what led to the decision, which organisations were involved, whether it arose out of a crisis or was something that had been discussed over a longer time span, details about the child’s prognosis at the time, who was providing regular support and care, who was involved in care planning, etc
- Collect information about who was involved / influential / important in the decision process – which professionals, which family members? Was their a care team involved in the decision – who was part of this team? How was the team involved in the decision process?

INFORMATION AND ADVICE

- Explore the types of information and advice parents were provided with to help them make a decision, and in what forms (written, verbal, online, etc)
- Discuss whether information and advice was provided to their child, how it was provided and in what forms, and whether it was appropriate for their level of ability and understanding
- Explore how professionals were involved in
 - giving information and advice to parents and children
 - supporting parents and children in the decision making process
- Did they understand the information and advice they were given and was it sufficient to make an informed decision?
- Did anyone take the time to discuss the information with parents and / or children and answer questions they may have had? Were they encouraged to ask questions and to make sure they understood the information provided?
- How many times was the decision discussed and with whom? Were parents given the time and support they needed to make a decision?
- Did parents or their child need more / less / different information, and if so from where / whom? Did they manage to find it?

- Did parents have a chance to talk to other parents whose child had a g-tube / ventilation? Did their child have a chance to talk to other children who had a g-tube / ventilation? How did this happen? Was it useful?
- What information / factors were important in helping them make a decision?

PARTICIPATION IN DECISION MAKING

- Collect information about their role / their child's role / professional's roles in the decision about technological support – how do they feel about their own and their child's level of participation?
- Were they actively encouraged to be involved in the decision process? If so, by whom and how?
- Was their child encouraged to be involved in the decision process? How was this achieved? Was it successful? Did it reflect their child's level of understanding and ability? Was their child's capacity for decision making assessed?
- Were parents asked about their preference concerning their own and their child's participation in decision making? Who by? Were efforts made to reflect these preferences, and if so how?
- Explore preference / opinions about their own and their child's participation in decisions about their child's care and treatment
- Explore preference / opinions about the role of health and other professionals in decision making for their child
- Who do parents feel made the final decision about technological support? Did they feel any pressure about making a decision? Did it feel like they were making a decision between different options?

DECISION OUTCOMES

- What difficulties did parents experience in deciding for their child to have a g-tube / ventilation? How could these difficulties have been avoided?
- What additional information and support did parents need after making a decision, and was this provided?
- Did parents feel prepared for managing the g-tube / assisted ventilation? Was information, training, support in place to help parents with this?
- If other parents in their position were having to make a decision about a g-tube or ventilation, what would they say to them / what advice would they give.

Appendix E – Interview Topic Guides: Young People

DECISION CONTEXT

- Collect information about the child and family – details of their illness, primary caregiver, family composition, who provides regular support and care
- Collect information about child's quality of life - likes and dislikes, things that are important to them, people important in their lives, whether they go to school
- Collect information about the decision to have a g-tube / ventilation – where and when the decision was made, which organisations were involved (hospice / hosp)
- Collect information about who was involved / influential / important in the decision process – which professionals, which family members?

INFORMATION AND ADVICE

- Discuss the sorts of information and support they were provided with, and in what forms (verbal, written, etc).
- Who provided information and support about getting a g-tube / ventilator – which professionals / family members / carers, etc
- Did they understand the information and advice they were given? Were they able to ask questions about the information, and to whom? How useful was the information they received?
- Did anyone talk to them about the decision / about having a gastrostomy or ventilation? – professionals / family members / parents etc
Were they encouraged to ask questions? Was it helpful to talk to others about it?
- Did they have a chance to talk to other children / young people who had a g-tube or used a ventilator. If so, was it useful?
- Did they need more / less / different information than what was provided, and if so from where / whom? Did they manage to find it?
- Which information / advice was really useful in helping them learn more about the g-tube / ventilator, and/or make a decision?

PARTICIPATION IN DECISION MAKING

- Explore their role in the decision to have a g-tube / ventilator – identify who helped them or encouraged them to become involved and whether they were happy about their level of involvement
- Who else was involved making the decision about g-tube / ventilation – explore the role of the other people involved (parents, professionals, other family members, etc)? Was this OK?

- Were they asked about how they wanted to be involved in the decision, and how they would like others to be involved in the decision? Did the decision process reflect these preferences?
- Explore children's preferences and opinions about their own participation and role in decisions relating to their care and treatment
- Explore children's preferences and opinions about other people's (parents, professionals, etc) participation and role in decisions relating to their care and treatment
- Who do they feel made the final decision about a g-tube / ventilation and was this OK?

DECISION OUTCOMES

- What additional information and support did children and young people feel they needed to help them learn about gastrostomy / ventilation?
- Did they feel prepared for the g-tube / assisted ventilation?
- What is their experience of having a g-tube or using a ventilator?
- If another child / young person was having to decide about a g-tube / ventilator, what would they say to him/her?

Appendix F – NHS Ethics Documentation: First Review Letter 23/06/08

Leeds (West) Research Ethics Committee

A/B Floor, Old Site
Leeds General Infirmary
Great George Street
Leeds
LS1 3EX

Telephone: 0113 3023181
Facsimile: 0113 3022863

23 June 2008

Ms Jo Nicholson
PhD Student
Social Policy Research Unit
University of York
Heslington
York
YO10 5DD

Dear Ms Nicholson

Full title of study: Decision processes in the use of technological support for children and young people with degenerative conditions
REC reference number: 08/H1307/74

The Research Ethics Committee reviewed the above application at the meeting held on 13 June 2008. Thank you for attending to discuss the study.

Ethical opinion

Whilst the Committee agreed that this is an important and worthwhile study using appropriate methodology, the members present decided they were unable to give a favourable ethical opinion of the research, for the following reasons:

1. Members had concerns about the lack of training and experience of the Chief Investigator to conduct the research in this very sensitive area. The Committee agreed that a person with clinical experience in supporting families in these circumstances should either conduct the interviews or be in attendance at the interviews to support the Chief Investigator.
2. Members were unhappy with the lack of immediate support available to both families and the Chief Investigator in a crisis situation.
3. The consent process was unclear in the case of conflict within the family with regard to taking part; members felt that in this situation such families should be excluded from the research.
4. Members thought the proposed arrangements for assessing capacity were inadequate and the assessment procedure should bear in mind the requirements of the Mental Capacity Act, including ensuring that an advance directive is not in place.

The Committee would like you to know that this was a very difficult decision, but we regret to inform you therefore that the application is not approved.

Mental Capacity Act 2005

The committee did not approve this research project for the purposes of the Mental Capacity Act 2005. The research may not be carried out on, or in relation to, a person who lacks capacity to consent to taking part in the project.

Options for further ethical review

You may submit a new application for ethical review, taking into account the Committee's concerns. You should enter details of this application at Question A55 on the application form and include a copy of this letter, together with a covering letter explaining what changes have been made from the previous application. The application should be booked through the Central Allocation System (CAS) and would be allocated for review in the normal way. You should let CAS know if you would like the application to be reviewed again by this Committee.

Alternatively, you may appeal against the decision of the Committee by seeking a second opinion on this application from another Research Ethics Committee. The appeal would be based on the application form and supporting documentation reviewed by this Committee, without amendment. If you wish to appeal, you should notify the Head Office of the National Research Ethics Service in writing within 90 days of the date of this letter. If the appeal is allowed, NRES will appoint another REC to give a second opinion within 60 days and will arrange for the second REC to be provided with a copy of the application, together with this letter and other relevant correspondence on the application. You will be notified of the arrangements for the meeting of the second REC and will be able to attend and/or make written representations if you wish to do so.

The relevant NRES contact point is:

Joan Kirkbride
Head of Operations, The North, Midlands & East of England
National Research Ethics Service (NRES)
National Patient Safety Agency
c/o Darlington Primary Care Trust
Dr Piper House
King Street
Darlington
Co Durham
DL3 6JL
joan.kirkbride@nres.npsa.nhs.uk

Documents reviewed

The documents reviewed at the meeting were:

Document	Version	Date
Application		09 May 2006
Investigator CV	Johanne Nicholson	09 May 2006
Investigator CV	Patricia Sloper	09 May 2006
Protocol	V1	31 March 2006
Covering Letter		09 May 2006
Participant Information Sheet: Young People - Short Text	V1	31 March 2006
Participant Information Sheet: Young People - Full Text	V1	31 March 2006

Participant Information Sheet: Bereaved Parents	V1	31 March 2008
Participant Information Sheet: Parents of Young People 18+	V1	31 March 2008
Participant Information Sheet: Parents of Children under 18	V1	31 March 2008
Participant Information Sheet: Key Professionals	V1	31 March 2008
Participant Information Sheet: Young People - Symbol	V1	31 March 2008
Participant Information Sheet: Young People - Simple Text	V1	31 March 2008
Participant Consent Form: Young People - Simple Text	V1	31 March 2008
Participant Consent Form: Young People - Full Text	V1	31 March 2008
Participant Consent Form: Parents/Guardians	V1	31 March 2008
Participant Consent Form: Key Professionals	V1	31 March 2008
Participant Consent Form: Young People - Symbol	V1	31 March 2008
Letters of invitation to participants: Parents of Children under 18	V1	31 March 2008
Letters of invitation to participants: Parents of Young People 18+	V1	31 March 2008
Letters of invitation to participants: Bereaved Parents	V1	31 March 2008
Letters of invitation to participants: Young People	V1	31 March 2008
Letters of invitation to participants: Key Professional	V1	31 March 2008
Statement of Indemnity Arrangements	Sue Final	10 April 2008
Topic Guides: Key Professionals	V1	31 March 2008
Topic Guides: Children and Young People	V1	31 March 2008
Topic Guides: Parents	V1	31 March 2008
Response Form: Young People and Parents	V1	31 March 2008
Response Form: Key Professionals	V1	31 March 2008
Response Form: Young Adults	V1	31 March 2008
Response Form: Parents	V1	31 March 2008
Response Form: Parents and Young People under 18	V1	31 March 2008
Cover Letter		23 May 2008
Cover Letter		22 May 2008

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Now that you have completed the application process please visit the National Research Ethics Website > After Review

Here you will find links to the following

- a) Providing feedback. You are invited to give your view of the service you have received from the National Research Ethics Service on the application procedure. If you wish to make your views known please use the feedback form available on the website.
- b) Re-submission/Appeal.

Appendix F – NHS Ethics Documentation: Second Review Letter 16/09/08

Newcastle & North Tyneside 1 Research Ethics Committee

Room 144
TEDCO Business Centre
Rolling Mill Road
Jarrow
NE32 3DT

Telephone: (0191) 4283561
Facsimile: (0191) 4283432

16 September 2008

Ms Jo Nicholson
Phd Student
Social Policy Research Unit
University of York
Heslington
York
YO10 5DD

Dear Ms Nicholson

Full title of study: Decision processes in the use of technological support for children and young people with degenerative conditions

REC reference number: 08/H0906/97

The Research Ethics Committee reviewed the above application at the meeting held on 09 September 2008. Thank you for attending to discuss the study.

Documents reviewed

The documents reviewed at the meeting were:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Application	Parts A+B	22 July 2008
Investigator CV	J Nicholson	21 July 2008
Investigator CV	P Sloper	22 July 2008
Protocol	v1	31 March 2008
Covering Letter	J Nicholson	22 July 2008
Letter from Sponsor	S Final	10 April 2008
Interview Schedules/Topic Guides	v1	31 March 2008
Interview Schedules/Topic Guides	v1	31 March 2008
Interview Schedules/Topic Guides	v1	31 March 2008
Letter of invitation to participant	v1	31 March 2008
Letter of invitation to participant	v1	31 March 2008
Letter of invitation to participant	v1	31 March 2008
Letter of invitation to participant	v1	31 March 2008
Letter of invitation to participant	v1	31 March 2008
Participant Information Sheet: Parent - child under 16	v1	31 March 2008
Participant Information Sheet: Parent - young people 16+	v1	31 March 2008
Participant Information Sheet: Bereaved parent	v1	31 March 2008

Ethics Response Letter

Participant Information Sheet: Young person - full text	v1	31 March 2008
Participant Information Sheet: Young person - short text	v1	31 March 2008
Participant Information Sheet: Young person - simple text	v1	31 March 2008
Participant Information Sheet: Symbol	not referenced	
Participant Information Sheet: Key professional	v1	31 March 2008
Participant Consent Form: Parent/guardian	v1	31 March 2008
Participant Consent Form: Young person - full text	v1	31 March 2008
Participant Consent Form: Young person - simple text	v1	31 March 2008
Participant Consent Form: Symbol	v1	31 March 2008
Participant Consent Form: Key professional	v1	31 March 2008
Unfavourable opinion letter	08A11307/74	23 June 2008
Response form - Parent/Young person under 16	v1	31 March 2008
Response form - Parent	v1	31 March 2008
Response form - young adult	v1	31 March 2008
Response form - key professional	v1	31 March 2008

Provisional opinion

The Committee wished to explore those situations which it felt could arise from the proposed interviews. The Committee considered that participants could be prompted to complain about some aspect of the care of their child as a result of discussions held during the study or may wish to use the interviews as an opportunity to make a complaint. You considered that such complaints would be unlikely but that they would be referred into the appropriate mechanism for the organisation affected. For example, the Hospice involved in the study has its own complaints mechanism. You indicated that you would take advice on the timing for referring any complaints back. The Committee suggested that a clear procedure for complaint handling should be in place at the start of the study. The Committee also wished to highlight that the research team will have access to and store information about the decision making process which may be relevant to litigation involving the organisations, families and/or staff participating. Such litigation would, in all likelihood, be wholly independent of the research study but the researchers could be directed to disclose any relevant information they hold in the course of such legal proceedings. Whilst accepting that this may be a remote possibility, the Committee suggested that the research team should recognise their potential role.

Previous ethical review of this application elsewhere had explored the research team's reaction to a parent consenting to participate but the child declining to participate. This Committee questioned whether the researchers would accept that it may be inappropriate to involve that parent since the child may not only not wish to actively participate but may not wish their situation to be discussed with the research team by the parent. You indicated that, in such circumstances, they would explore the reason for any child withholding consent and make a decision on the inclusion of the parent on a case by case basis. They will also have input from the Hospice involved in the recruitment process and this may provide additional information about the child and any reason for declining to participate. The Committee acknowledged that the role of the Hospice as a 'gatekeeper' or filter could provide valuable additional guidance in such situations.

The Committee expressed concern in respect of the level of some of the language used in the participant information documentation - you indicated that a consultation group has been involved in the design of the study and that you can consult this group about the suitability of the language.

The Committee expressed concern in respect of the information documentation to be used with bereaved parents -this should not simply employ the same phraseology to be used with other groups - alternative and carefully worded documentation should be devised which acknowledges their circumstances. You accepted this point and agreed to submit revised documentation.

The Committee questioned why potential participants would be asked at an early stage in the recruitment process about the nature of the clinical intervention which had applied to their child. It was suggested that this could be established at the interview stage or from the recruiting organisation. You indicated that the information will assist in structuring the sample but that the information can be obtained at the interview stage.

The Committee suggested that the consent documentation should make clearer that information provided at the consent stage would be securely stored while still required and will be destroyed at an early stage when no longer required - this point was accepted.

It was suggested that the 'simple text' version of the information documentation for younger persons should contain a little more detail of the study.

The Committee also made the following points:

- the information for participants should make clear that if, during the course of study participation, it becomes apparent that advice should be sought in respect of potential litigation or professional standards, study participation may be suspended;
- there is no indication that there is an appropriate 'lone worker' policy available and in place;
- the amount of the ESRC funding is not stated.

The Committee is unable to give an ethical opinion on the basis of the information and documentation received so far. Before confirming its opinion, the Committee requests that you provide the further information set out below.

The Committee delegated authority to confirm its final opinion on the application to the Chair.

Further information or clarification required

The Committee suggested that a clear procedure for complaint handling should be in place at the start of the study.

The Committee expressed concern in respect of the level of some of the language used in the participant information documentation - you indicated that a consultation group has been involved in the design of the study and that you can consult this group about the suitability of the language.

The Committee expressed concern in respect of the information documentation to be used with bereaved parents -this should not simply employ the same phraseology to be used with other groups - alternative and carefully worded documentation should be devised which acknowledges their circumstances. You accepted this point and agreed to submit revised documentation.

The Committee questioned why potential participants would be asked at an early stage in the recruitment process about the nature of the clinical intervention which had applied to their child. It was suggested that this could be established at the interview stage or from the

recruiting organisation. You indicated that the information will assist in structuring the sample but that the information can be obtained at the interview stage.

The Committee suggested that the consent documentation should make clearer that information provided at the consent stage would be securely stored while still required and will be destroyed at an early stage when no longer required - this point was accepted.

It was suggested that the 'simple text' version of the information documentation for younger persons should contain a little more detail of the study.

The Committee also made the following points:

- the information for participants should make clear that if, during the course of study participation, it becomes apparent that advice should be sought in respect of potential litigation or professional standards, study participation may be suspended;
- there is no indication that there is an appropriate 'lone worker' policy available and in place;
- the amount of the ESRC funding is not stated.

When submitting your response to the Committee, please send revised documentation where appropriate underlining or otherwise highlighting the changes you have made and giving revised version numbers and dates.

The Committee will confirm the final ethical opinion within a maximum of 60 days from the date of initial receipt of the application, excluding the time taken by you to respond fully to the above points. A response should be submitted by no later than 14 January 2009.

Ethical review of research sites

The Committee agreed that all sites in this study should be exempt from site-specific assessment (SSA). There is no need to submit the Site-Specific Information Form to any Research Ethics Committee. However, all researchers and local research collaborators who intend to participate in this study at NHS sites should seek approval from the R&D office for the relevant care organisation.

Membership of the Committee

The members of the Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

08/H0906/97	Please quote this number on all correspondence
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Yours sincerely

Appendix F – NHS Ethics Documentation: Final Approval Letter 05/11/08



National Research Ethics Service

Newcastle & North Tyneside 1 Research Ethics Committee

Newcastle Dental School
Room G14
Dental School
Framlington Place
Newcastle
NE2 4BW

Telephone: 0191 428 3561
Facsimile: 0191 428 3432

05 November 2008

Ms Jo Nicholson
PhD Student
Social Policy Research Unit
University of York
Heslington
York
YO10 5DD

Dear Ms Nicholson

Full title of study: Decision processes in the use of technological support for children and young people with degenerative conditions
REC reference number: 08/H0906/97

Thank you for your letter of 30 October 2008, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Ethical review of research sites

The Committee has designated this study as exempt from site-specific assessment (SSA). The favourable opinion for the study applies to all sites involved in the research. There is no requirement for other Local Research Ethics Committees to be informed or SSA to be carried out at each site.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission at NHS sites ("R&D approval") should be obtained from the relevant care organisation(s) in accordance with NHS research governance arrangements.

This Research Ethics Committee is an advisory committee to North East Strategic Health Authority
The National Research Ethics Service (NRES) represents the NRES Directorate within
the National Patient Safety Agency and Research Ethics Committees in England

Guidance on applying for NHS permission is available in the Integrated Research Application System or at <http://www.rdforum.nhs.uk>.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Covering Letter	J Nicholson	22 July 2008
Unfavourable opinion letter	08/H1307/74	23 June 2008
Application	Parts A+B	22 July 2008
Letter from Sponsor	S Final	10 April 2008
Protocol	v1	31 March 2008
Investigator CV	J Nicholson	21 July 2008
Investigator CV	P Sloper	22 July 2008
Participant Information Sheet: Symbol	not referenced	
Participant Consent Form: Symbol	v1	31 March 2008
Interview Schedules/Topic Guides	v1	31 March 2008
Interview Schedules/Topic Guides	v1	31 March 2008
Interview Schedules/Topic Guides	v1	31 March 2008
Response to Request for Further Information	Johanna Nicholson	30 October 2008
Participant Information Sheet: Information Leaflet	Bereaved Parents - Version 2	30 September 2008
Participant Information Sheet: Information Leaflet	Parents of Young People 16+ - Version 2	30 September 2008
Participant Information Sheet: Information Leaflet	Parents of Children under 16 - Version 2	30 September 2008
Participant Information Sheet: Information Leaflet	Key Professionals - Version 2	30 September 2008
Permission Form	Young People and Parents - Version 2	30 September 2008
Response Form	Key Professionals - Version 2	30 September 2008
Response Form	Young Adults - Version 2	30 September 2008
Response Form	Parents - Version 2	30 September 2008
Response Form	Parents and young people under 16 - Version 2	30 September 2008
Participant Consent Form: Consent Form	Parents and Young People under 16 - Version 2	30 September 2008
Participant Consent Form: Consent Form	Young People (simple text) - Version 2	30 September 2008
Participant Consent Form: Consent Form	Young People (full text) - Version 2	30 September 2008
Participant Consent Form: Consent Form	Young People Age 18 and over 18 - Version 1	30 September 2008
Participant Consent Form: Consent Form	Parents/guardians - Version 2	30 September 2008
Safety on Fieldwork		31 March 2008
Participant Information Sheet: Information Leaflet	Young People (full text) - Version 2	30 September 2008
Participant Information Sheet: Information leaflet	Young People (short text) - Version 2	30 September 2008
Participant Information Sheet: Information Leaflet	Young People (simple text) - Version 2	30 September 2008
Letter of invitation to participant	Bereaved Parents - Version 2	30 September 2008

This Research Ethics Committee is an advisory committee to North East Strategic Health Authority
The National Research Ethics Service (NRES) represents the NRES Directorate within
the National Patient Safety Agency and Research Ethics Committees in England

Letter of invitation to participant	Parents of Children Under 16 - Version 2	30 September 2008
Letter of invitation to participant	Parents of Young People 16+ - Version 2	30 September 2008
Letter of invitation to participant	Young Adults - Version 2	30 September 2008
Letter of invitation to participant	Key Professionals - Version 2	30 September 2008

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Now that you have completed the application process please visit the National Research Ethics Website > After Review

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

08/H0906/97

Please quote this number on all correspondence

With the Committee's best wishes for the success of this project.

Yours sincerely

A. Taylor
 pp Mr Chris Turnock
 Chair

Email: Anne.taylor7@nhs.net

Enclosures: "After ethical review – guidance for researchers"

This Research Ethics Committee is an advisory committee to North East Strategic Health Authority
 The National Research Ethics Service (NRES) represents the NRES Directorate within
 the National Patient Safety Agency and Research Ethics Committees in England

Appendix G – Support Leaflet for Participants



If you would like regular updates on the project or have any questions about the project, please contact me:

Name: Jo Nicholson
Telephone: 01904 321969
Email: jn123@york.ac.uk
Address: Social Policy Research Unit
University of York
York YO10 5DD
Website: www.york.ac.uk/spru

Research project about gastrostomy or assisted ventilation for children and young people

SUPPORT AND ADVICE

If, after your interview, you feel that you would like some support or advice, this leaflet contains some useful links and resources available to families and their children. Alternatively, please contact me on 01904 321969 or at jn123@york.ac.uk.

Thank you for taking part in this study.

Jo Nicholson

THE UNIVERSITY *of York*

PALS - Patient Advice and Liaison Service

PALS has been set up by the NHS to provide patients and their families advice and assistance about their care and treatment, and also offer information about services they wish to access.

PALS can also act on your behalf by talking to staff, managers, and other relevant organisations to address your concerns. They also have access to local and national-based support agencies.

Your local PALS:

Address:

Address:

Tel:

Email:

ICAS - Independent Complaints Advocacy Service

ICAS provides advice and support to people who wish to make a complaint about the treatment or care they, or a family member has received from the NHS.

They will provide you with information about the NHS complaints process, and offer you advice and guidance. They can also support you through the complaints process.

The ICAS service is free, independent and confidential.

Your local ICAS:

Address:

Address:

Tel:

Email:

Support from Martin House Children's Hospice

If you would prefer to talk to some-one at Martin House, who initially invited you to take part in the project, please contact Jan Aldridge, who is supervising Jo's work, and is based at Martin House. She is happy to answer any questions you may have, or assist you in finding the right support.

Tel: 01937 845045

Email: jan@martinhouse.org.uk

Appendix H – Thematic Framework for Data Analysis

1	The Decision Context
1.1	The Child
1.2	The Family
1.3	The Decision
1.4	Background to Decision
1.5	Summary of need for technology
1.6	What else was the decision about?
1.7	Time Period
1.8	The Care Team
1.9	Who was involved?
1.10	Key Professional
1.11	Outcomes
1.12	Other consequences

2	Decision Factors
2.1	Choice
2.2	Medical Need
2.3	Invasiveness
2.4	Risks
2.5	Quality of Life
2.6	Treatment philosophy
2.7	In whose best interests?
2.8	Uncertainty – prognostic and outcomes
2.9	Resilience of the child (Close Calls)
2.10	Other

3	The Decision Process
3.1	The recommendation
3.2	Information Exchange – Flow
3.3	Information Exchange – Direction
3.4	Information Exchange – Type
3.5	Information Exchange – Amount
3.6	Using and interpreting information
3.7	Deliberation
3.8	The Second Opinion
3.9	Giving consent / refusal
3.10	Decisional Control
3.11	Other

4	Parents as Decision Makers
4.1	Gaining Control
4.2	Doing What it Takes
4.3	She's My Child
4.4	Parental Instinct
4.5	Being Rational
4.6	Decisional Responsibility
4.7	Being Informed
4.8	Other

5	Making Decisions with Professionals
5.1	The Triadic Relationship
5.2	Professional Parent Interactions
5.3	Professional Expertise
5.4	Incorporating the bigger picture
5.5	The good ones – trust, empathy, compassion
5.6	Professional Bias
5.7	Other

6	The System
6.1	Trust in the system
6.2	The Rules and Red Tape
6.3	Fragmentation of services and expertise
6.4	Hospital Experience
6.5	Having some-one on the inside
6.6	Continuity of care
6.7	The role of specialist services
6.8	Transition to adult services
6.9	Other

7	The Child's Expanding Role
7.1	Developing capacity for decisions
7.2	Parents as teachers for their child
7.3	Informing the child
7.4	Managing fear
7.5	Condition / prognostic awareness
7.6	Other

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