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An exploration of the experiences of children affected by life limiting conditions and their families

by

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Dedicated to my mother

Sybil Mary Magdeline Menezes

4th August 1925 – 1st June 2005

To know in a moment that being together
Is such a fragile thing
Makes you look again at
Yourself, your life
And who you are

UNIVERSITY OF SOUTHAMPTON
<u>ABSTRACT</u>
FACULTY OF MEDICINE, HEALTH AND LIFE SCIENCES
HEALTH SCIENCES GRADUATE SCHOOL
SCHOOL OF NURSING AND MIDWIFERY
<u>Doctor of Philosophy</u>

AN EXPLORATION OF THE EXPERIENCES OF CHILDREN AFFECTED BY LIFE LIMITING CONDITIONS AND THEIR FAMILIES

By Antoinette Maria Menezes

This thesis provides an account of a qualitative study that set out to understand more about the needs and experiences of children affected by life limiting conditions and their families. Participants included eleven children, their parents and siblings from ten families (39 participants in all).

The theoretical approach is drawn towards interpretism and constructionism. Case study provided the overarching strategy and aspects of the design were drawn from Grounded Theory. The methods combined participant observation, interviews and an invitation for young participants, especially, to use their own artwork and photographs to help them explain their day-to-day experiences.

Findings are presented initially through five conceptual cases (all based on the life stories of more than one child). The study identified recurring 'moments of realisation' in the children's life stories and revealed that these were the times when families recognised the threat to the child's life most clearly and needed to communicate with each other but struggled to do so. It is hoped that the conceptual cases and the concept of 'moments of realisation' will provide professionals with new ways to think about the needs and experiences of life limited children and their families. This concept challenges notions about 'one moment' to talk to the child about the life limiting nature of their illness and the somewhat linear notions of illness and dying trajectory.

Participants in the research used a range of indirect topics to talk about the child's illness and dying through the research process. These are used to suggest new ideas for helping life limited children and their families to talk to each other about the child's illness and it's consequences.

The findings suggest that children affected by life limiting conditions gradually integrate information about their illness and short life expectancy into their sense of self and their own life story. The children challenged concepts of biographical disruption because they demonstrated such capacity to incorporate the illness into their life stories and live life to the full.

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Introduction

Research problem and rationale for the study

This thesis describes research that focused on the experiences of eleven children (from ten families) diagnosed with conditions predicted to limit their life expectancy to childhood or only early adult years. Parents and siblings from these ten families were also vital contributors and there were 39 participants in total. Hardly any research to date has examined the child and family's perception of this rare but overwhelming experience.

The purpose of this study was to hear from 'life-limited' children about their day-to-day encounters and to bring the stories of the children and their families side-by-side. The aim was to understand more about the child's perspective as the member of a family and in the context of family life.

What faces life limited children and their families is the stark prospect of the child's death at some unknown point in the future. The path ahead may have little or no light on it (given the rare nature of some children's medical diagnoses). Deterioration in the child's health can follow many patterns but is likely to be protracted and may include survival from one or many critical episodes. The time from diagnosis to the child's death may span many years and yet little is known about the needs and experiences of the child and family members or how their needs might be met. Where services to support children with life limiting conditions and their families do exist, the networks of provision and communication are complicated and most are funded by the voluntary sector. As a result families often cope alone, perhaps for many years, with all the emotional, social and financial costs that this entails.

Study aims

In broad terms this study aimed to further understanding about the experiences of children living with life-limiting conditions and the experiences of their families; to uncover more about the nature of this experience, as it is perceived internally (by the children and their families) and how outsiders interpret and respond to this rare (not just unusual) situation. The following questions arose:

1. How do families face the dilemma of whether or how to be open about the life limiting nature of the child's condition within the family?
2. What is the impact of having a life limiting condition on a child's developing sense of self?
3. Do children who are unable to communicate verbally have active and/or passive roles in shaping their identities?
4. What are the current roles of support services such as children's hospices? What are the challenges and opportunities for developing support that matches the needs of life limited children and their families more closely?

Chapter One

1 Background and context

1.1 Theoretical approach

The experience of life limiting illness from the point of view of affected children has not been researched and is not well understood. I approached the research questions from the stance of interpretism and social constructionism for a number of reasons. The research questions presupposed the ability of children to represent themselves through research. It was presumed that this might be achieved by meeting the children and their families, in the general contexts of their lives (in their own homes and schools), hearing from them in their own words (where they could communicate verbally) or by being present, observing and participating in events from their daily lives and sharing their encounters.

From the interpretive perspective knowledge is not objective, inherent and waiting to be discovered but constructed by people who are constantly interpreting, developing ideas and meanings and passing them on or re-evaluating and reconstructing new concepts. Interpretists tend to view knowledge as subjective, assuming that understanding is drawn from experience and personal perceptions. Therefore, if the epistemological stance of the researcher is based on the notion that knowledge is generated through a complex partnership between subjects and objects to construct social meaning, which is personal and unique to individuals, then the researcher will tend to seek involvement with those who participate in their research, valuing the uniqueness of each participant and differences within the group as much as any similarities.

Max Weber's theory that society was shaped by individual reactions to it suggests that people make and alter social order. Constructionism is an extension of this theory, arguing that all social phenomena are 'constructed'. They do not exist without recognition and response, so that by another society they may not be constructed as real or could be interpreted differently (Morrall, 2001). Burr (2000) described four principles of the social constructionist approach:

1. A critical stance towards the idea that the nature of the world can only be uncovered by observation and measurement (positivism).
2. An acceptance that we understand the world in ways that are relative to time and place, that are historically and culturally specific.
3. An acceptance that knowledge is constructed through social interactions, especially language. That language is a pre-condition of thought not just an expression of it.
4. The understanding that socially constructed theories tend to invite response.

Social constructionists bind truth and knowledge to perspective, supposing that meaning is related to context. Social constructionists place emphasis not on individuals constructing meaning in isolation but rather on the collective processes of communication and language, social interaction and all aspects of social context (environmental, historical and cultural). From the view of social constructionism predominant ideas come from language and communication between people and are most powerful in context and might be meaningless or less influential at a different place or time.

People construct knowledge together and research guided by social constructionism might set out to understand more about the way social groups construct meaning around notions such as 'childhood' or 'illness' and 'death in childhood'. The balance of power in social construction is important because inevitably some social groups have greater influence than others. Involving children in social constructionist research might also reveal how children (who have a relatively less powerful place in society than adults) might choose to represent themselves and their lives and reveal how children might construct their own meanings about ideas such as 'childhood', 'illness' or 'death and dying in childhood'.

This research was embedded in interpretism and social constructionism because it set out to understand the subjective interpretations of participants (children with life limiting conditions and their families). The aim of the design was to create a framework for the children, their families and myself to work in partnership. This was intended to be a joint venture in which my own function was to keep the children and families focused on the aims of the study.

1.2 A personal account

Within qualitative research it is acknowledged that the identity, values and beliefs of the researcher cannot be entirely eliminated from the research process. Therefore in contrast to the ambitions of the positivistic approach, the interpretist attempts to explore their subjective position or at least bring it to the fore. Some years ago I was senior staff nurse on a ward for people with respiratory problems. Many of the patients were young people with cystic fibrosis who were admitted on repeated occasions. One Monday afternoon I returned to work after my weekend off and was confronted by a teenage girl who suffered from cystic fibrosis. She demanded, 'Where were you all weekend?' I resented her tone and the implication of her question, did she think I didn't deserve a break … I asked, 'Why?'

She explained, 'I was admitted on Friday and there was no room for me here so I got put on another ward. No one knew me. I was really sick and they all thought I was dying so they put me in a side room, out of the way. All weekend no one came in unless they had to.' I sat next to her not knowing what to say except 'Sorry'. She looked at me for a time and then put her hand on my shoulder, 'I just needed people I knew. I knew I might die and I didn't want to be alone. The nurses didn't know what to say to me … so they said nothing, unless they had to 'do things' like sort out the drip …' She was 16 and she died within months.

This moment stayed with me for many years. At the time I felt inadequate and frustrated with the system that I was part of but felt powerless to change. I was left with questions about how services and individuals might respond better to the needs of young people who may only have a short time to live. This brief moment with a dying 16-year-old girl began my journey to this research. There were many steps along the path including an MSc study in which I explored the experiences of parents whose children suffered brain injury after cardiac surgery. However I did not yet have the courage to talk to the children directly. Over the next two or three years, being part of a new and emerging children's hospice service and undertaking training in 'How to involve children in research' (held at

York University), developed in me a belief that children are insightful about their own lives and can actively participate in research.

I began this research as a nurse concerned about the experiences of an adolescent in my care. I acknowledge that being part of a children's hospice service has inspired my approach to this research and permitted me contact with a group of children and families who would be difficult to recruit any other way. Since each child and their family require a package of care, children's hospices are part of a broader network of palliative care services for children. Therefore I have sought to understand more about the needs and experiences of life limited children and their families, not from the perspective of one service but through the children and families' explanations of their own lives, and day-to-day experiences.

I decided at the outset of the study to be open with parents that I was the primary carer for a close family member, above all in case I had to cancel planned visits with the children. Most parents just accepted my simple explanation and told me to just let them know. Two mothers and one adolescent asked who I cared for and enquired about my mother whenever we met. Although the stories of some families mirrored my own experiences I did not discuss this with participants in case it changed the focus of their interviews from their own priorities to mine. I had access to an independent counsellor throughout the study and he provided me with invaluable support for all personal and professional issues that arose. I also used a reflective diary to capture and explore my own experience of the study.

At the start of the study I worried whether any child would confide in me at all. My natural instinct was to be playful with children. Although I have no children of my own, the children of family and friends are people whose company I enjoy and we have lots of fun together. Being a 'researcher' trying to work with children was different and uncomfortable for me to begin with. However I made an early entry in my reflective diary to make no judgements about work with the first child until our third meeting. This was good advice to myself.

1.3 Defining terms

In 1993 it was suggested that understanding about needs and care requirements for children with life limiting conditions could be held back by the absence of agreed definitions for even the most basic terms - for example, 'child' (Sutherland et al, 1993).

In keeping with other relevant policy and service development guidelines, children will be defined here, as young people from birth to 18 years of age (ACT and RCPCH, 2003; British Government, 1989). The children at the centre of this research represent a complex group who suffer from a wide range of medical conditions all with different incidence patterns, symptoms and consequences.

> *"A clear understanding of the client group is required since children in need of palliative care are likely to cross other classifications used by health (for example, children with disabilities or long-term illness) social services (for example, children with special needs) and education (for example, children with special educational needs)." (ACT and RCPCH, 2003 p11)*

There is little consistency within the emerging literature about how to refer to children living with shortened life expectancy over months or years (Crossley et al, 1994). An early analysis of the work of a children's hospice (Burne et al, 1984) described the children's conditions as: terminal illness; progressive and incurable; and very severe handicap. In the last decade authors have sometimes defined this group of children as life threatened and likely to die prematurely from non-malignant disease (Lenton et al, 2001) or as children with neurodegenerative life threatening illnesses (Steele, 2000). Others (Nash, 1998; While et al, 1996a) refer instead to children suffering from life limiting conditions who are likely to die in childhood (before the age of 19). Some efforts have been made to disentangle the differences between 'life threatening' and 'life limiting' conditions. Sutherland et al (1993) suggested that life threatening conditions are those for which medical cure may be possible but life limiting conditions are those for which no curative treatment currently exists. However Lenton et al (2001) found little distinction between use of these terms and argued that individuals can oscillate between chronic illness or disability which becomes a threat to their lives, especially at times of acute exacerbation.

In terms of general language, the word 'threatened' indicates ominous signs of a source of harm or danger. It is interesting to note that the opposite of threatened is reassured. Anything that is threatening is intimidating and imperils the individual under threat, however the outcome of threat is never certain. When authors write about children with 'life threatening' conditions they indicate an illness that places the child's life in jeopardy but might be cured. Frequently potential treatment is expressed in language reserved for combat (for example, 'battle', 'brave fight', 'not giving in') implying an active role for the 'threatened' and their 'allies' (Dixon-Woods et al, 2003). These notions are common in the general media and can be found in medical literature (Davies et al, 2004; Dixon-Woods et al, 2003).

In everyday use the word 'limited' is more complex. It refers to bounded phenomena, restricted in some way or having a parameter (boundary or edge). The opposite of limited is limitless or infinite. In referring to medical diagnoses as 'life limiting' it appears that reference is made to the unexpected and untimely border around the child's life expectancy; a life that is likely to be shortened but often with immense uncertainty about when. The language of 'conflict' is not so evident in the media or medical literature perhaps because the 'fight' seems futile when cure is doubtful. We might resist a threat but a boundary is inevitable, even if it is invisible for a time.

From a social constructionist perspective, language is relevant because it plays a role in the wider construction of meaning. What does it mean socially to hear that a child is 'life limited' or a group of children suffer from 'life threatening illness'? What do these terms mean to the children and families? How do services build systems of support for 'life limited' children and their families?

ACT and RCPCH (2003) take 'life limiting conditions' as the overarching term and delineate it into four broad groups:

a. Life threatening conditions for which curative treatment may be feasible but can fail for example, Cancer.
b. Conditions where there may be long periods of intensive treatment but premature death is still possible for example, Muscular dystrophy

c. Progressive conditions without curative treatment options, where treatment is palliative but may extend over many years for example, Batten's Disease.
d. Conditions with severe neurological disability, which may cause weakness and susceptibility to health complications for example, severe multiple disabilities following brain or spinal cord injury.

(ACT and RCPCH, 1997 p7)

The terminology used throughout this thesis follows that given by ACT and RCPCH, (1997). 'Life limiting' is used as the overarching term for conditions that shorten the life expectancy of affected children. However, terms used by other authors are drawn into the literature review so that the essence of original research is not diluted and to maintain the integrity of the review.

1.4 Epidemiological data

Information about the number of children suffering from life limiting conditions is fragmented and uncertain, still failing to clarify what Baum (1994) called 'the magnitude of the problem'. Broadly deaths in childhood can be divided into those which are sudden - for example from road traffic accidents, and those which are not immediate but inevitable - for example, from Batten's Disease (Baum, 1994). Difference between these equally devastating experiences, lies in the cumulative pressures on families of facing this prospect on a day-to-day basis and perhaps in opportunities for the child and their family to prepare and to be supported in this process. Mortality rates are available in the UK (British Government, 2003) but only show the number of children who die at any given time. They do not account for children living with conditions that limit their life expectancy.

In a series of epidemiological studies based on the medical records of children with 'complex chronic conditions' who died in Washington, Feudtner and colleagues (Feudtner et al. 2002; Feudtner, Christakis, and Connell 2000; Feudtner, DiGiuseppe, and Neff 2003; Feudtner, Silveira, and Christakis 2002) illustrated where these children died and the complexity of their needs. The authors classified nine major categories of complex chronic conditions: cardiovascular, neuromuscular, malignancy, respiratory, renal, metabolic, gastrointestinal, haematologic/immunologic, and other congenital/genetic conditions. It was found that children who died of complex chronic conditions suffered from multiple medical conditions and 44% had a diagnosis representing one major category, 13% had diagnoses representing two major categories and 4% had diagnoses representing three or more major categories (Feudtner et al, 2002). Feudtner, DiGiuseppe, and Neff (2003) found that unlike infants, children and young adults who died of chronic conditions other than leukemia lived outside of the hospital for most of the last year of their life and were more likely to die at home (Feudtner et al, 2002). Despite arguing for a non-categorical approach to the development of support, Feudtner et al (2002) also highlighted the need for flexibility so that individual differences would still be catered for.

In the UK While et al., (1996a) listed the many conditions that limit life expectancy in childhood. However, prevalence data existed for very few of the more common conditions - for example, cancer and duchenne's muscular dystrophy (ACT and RCPCH, 2003). Some conditions are so rare that there may be only one affected child in this country or across the world. Even if robust information about prevalence were available, issues related to life expectancy would still be complicated, often depending on the vulnerability or vigour of each child. Therefore the exact number of children in the UK with life-limiting conditions is not known and the issues are complex. Figures tend to converge on 10 per 10,000 children aged 0-19 per annum (ACT and RCPCH, 2003) and it is estimated that there are between 15-20,000 children in the UK with conditions that mean they will not live to reach adulthood, (ACH, 2000).

Lenton et al (2001) described morbidity in 93 children suffering from non-malignant life threatening conditions. It was found that of the children assessed most suffered from multiple disabilities: 64% had problems with mobility; 57% had problems with manipulation and hand function; 45% had some difficulty expressing or understanding spoken language; 33% had hearing impairments; and 25% suffered visual impairment. This study illustrates that many children suffering from life limiting conditions have complex physical needs and require constant and complete care. Others suffer progressive deterioration, so that skills and abilities dissipate whilst care needs increase. Some of the children are able to attend mainstream schools whilst others have learning difficulties and require individual educational support. Children across this wide range of needs and ages need to be represented in research to set in motion the process of capturing information that reflects this varied group. Then evolution of services catering for these children and their families can begin to be based on evidence that reflects the extent of their diversity.

1.5 Service provision

Thornes (1990) described the key findings of the working party on the 'care of dying children and their families' set up by the National Association of Health Authorities (NAHA), the King's Fund and British Paediatric Association in 1987. The distinct needs of life limited children were not clear and the word 'hospice' seemed to generate a misconception amongst the general public of children dying of cancer who needed facilities like those provided for adults (Thornes, 1990).

In 1996/97 the Department of Health commissioned the studies of While et al (1996a & b – both studies are described in detail in the appended Table of Evidence) and an evaluation of the pilot project programme for children with life threatening illnesses (DoH, 1998). Recommendations included: better coordination of care in each district; inclusion of children's community nurses and social workers in palliative care teams; and opportunities for children and families to have respite breaks. Goldman (1994) argued that support available at that time was inadequate and distributed unevenly. Noting that much of the available support was provided by charities, Goldman (1994) urged Health Authorities to give priority and identify funding to fill the gaps in provision of palliative care for children.

> *"Palliative care for children and young people with life limiting conditions is an active and total approach to care, embracing physical, emotional, social and spiritual elements. It focuses on quality of life for the child and support for the family and includes the management of distressing symptoms, provision of respite and care through death and bereavement. It is provided for children for whom curative treatment is no longer the main focus of care and may extend over many years." (ACT and RCPC, 2003 p6)*

Palliative care for children is described (ACT and RCPCH, 2003) as special in the following ways:

- Fewer children than adults die
- Death in childhood can be caused by many extremely rare conditions
- Children may need palliative care for a few days, months or many years
- Many of the illnesses are familial and genetic counselling may be needed
- Care should include the whole family who are all affected by the child's illness
- Physical, emotional and cognitive development is reflected in children's communication skills and the ways that they understand their illness and death
- Play is vital to children and education is a legal entitlement

Care for life limited children and their families is inter-disciplinary and combines elements of statutory and voluntary sector provision. In the community, palliative care for children necessitates partnership and collaboration between paediatricians, community children's nurses, children's hospices, social workers, psychologists, occupational therapists, physiotherapists and speech and language therapists (ACT and RCPCH, 2003). It is argued therefore that,

> *"Well co-ordinated support between specialist centres and local teams is needed to prevent families feeling isolated." (ACT and RCPCH, 2003 p10)*

The first children's hospice in the world was opened in Oxford (Helen House) in 1982. To date 32 children's hospices have opened across England, Wales, Scotland and Northern Ireland, and ten more are planned. Children's hospices are independently run charity funded bodies. Children's hospices operate on various models, sometimes providing community based care as well as opportunities for children and families to take respite breaks at purpose built facilities. Care and support is available from the moment of diagnosis to the child's death and for the family through bereavement.

Stein et al (1989) examined family perceptions of the care provided by one children's hospice using semi-structured interviews and self-report questionnaires. It was found that parents often self referred to the hospice and valued the non-hospital environment, the emotional support and time to talk with staff. Families valued staff experience especially if their child had a rare condition (Stein et al, 1989). Symptom control was important. Although parents had reservations about exposure of their child to other sick and dying children, they found it helpful to meet other families with similar experiences to their own (Stein et al, 1989). Support for the whole family including siblings was very important to the families studied.

An independent evaluation of a children's hospice (Phillips and Burt, 1999) found that at first the idea of referral to a children's hospice concerned some families who feared it signified that their child was dying. The researchers called for better information at a national level. Phillips and Burt (1999) interviewed 28 children and family members. Young people highlighted that hospice services provided for them sometimes failed to offer written information for children or young people of any age and argued that this was an important consideration (Phillips and Burt, 1999). The researchers included children who communicated non-verbally by involving individuals who knew the children well (usually parents) and showing the children photographs of the hospice and staff to observe their reactions to simple questions. It was suggested that with extra time and experience more might have been done to include these children and understand their experiences (Phillips and Burt, 1999).

A survey of carers whose children were referred to a community based palliative care service included the administration of questionnaires at referral and six months later (Horrocks, Somerset, and Salisbury 2003). The results demonstrated that before using the service nearly half the children were rated by their main carer as experiencing considerable distress. Perceived problems were frequently due to emotional or practical care needs rather than the child's physical illness. Carers reported that their information needs, and the child's needs for nursing care improved with support from the service. Distress ratings reduced slightly after families began to use the service but there was no decrease in needs for equipment or respite (Horrocks, Somerset, and Salisbury 2003).

The study of Robinson and Jackson (1999) provided a description of children's hospice use and provision. The average age of the children was 9.7 years and one fifth had severely limited life expectancy. One third of families were lone parents and 12% came from minority ethnic backgrounds. Families were said to value the facilities, environment and care provided. For one third of families the hospice was their only source of respite and the others used one other facility. On average children stayed at the hospice for 20 days/year. The researchers were critical of the lack of planned activities for children with multiple impairments on the three days they spent at four children's hospices (Robinson and Jackson, 1999).

Notably absent from most of these studies are the voices of the children using the services. Article 12 of the United Nations Convention on the Rights of the Child (1989) states that children have a right to be consulted on matters of concern to themselves. Article 23 states that a mentally or physically disabled child should enjoy a full and decent life, in conditions that ensure dignity, promote self-reliance and facilitate the child's active participation in the community.

1.6 Structure of the thesis

Chapter two describes a review of the literature, beginning with an introduction that explores the theoretical implications of involving life limited children in research. Particular attention is paid to selected theories of child development relevant to: personality and identity development, children's developing understanding of death, potential for the growth of all children through social interactions, and the complex social systems within which children live. Although theories of child development provide structures for understanding how most children change as they grow, it is argued that they fail to account for differences in individuals.

The literature review illustrates how little research is available to uncover what it is like to live and grow up as a child affected by a condition that limits life expectancy to childhood (from the child's point of view). Available studies tended to focus on the experience of cancer in childhood but rarely included the child's perspective of living with shortened life expectancy over months or years.

Children suffering from the wide range of life limiting conditions, particularly those with complex disabilities and communication difficulties are barely represented. The voices of life limited children and their families about their day-to-day experiences are largely missing from research. Finding ways to hear from this group with mixed skills and abilities might provide insights relevant to studies of other vulnerable social groups.

The three central arguments of the review are firstly, that experience may enhance children's concepts of death in spite of their chronological age and therefore be very relevant to our understanding of dying children. Secondly, children suffering from life limiting conditions need information about their diagnosis, treatment and prognosis. However, there is limited evidence based advice for adults about how to communicate with children about their own dying and virtually no advice about whether, when or how to raise these topics with children who may have months or years to live. Thirdly, the review suggests that the child's illness impacts on the whole family and families manage by understanding and reacting to overarching patterns in the child's illness. Therefore, families want professionals to understand the wider picture of their lives, although the importance of including whole families is rarely considered in research design.

Chapter three describes the methodology, highlighting some key issues about involving children in research from the current ethical debates to consideration for the links between children and their families. Special attention is paid to the inclusion of children who communicate non-verbally. It is suggested that the challenge for researchers is to find ways to include all children with experiences relevant to the research question, from the youngest to the most disabled.

Case study was adopted as the overarching strategy and the design draws on grounded theory methodology. A case study approach is considered pertinent to the investigation of people centred problems, focusing on contemporary phenomenon in real-life context, when the boundaries between phenomenon and context are not clearly evident. Yin (1994) argued that the distinguishing strength of case study is the opportunity it offers to include a wide range of evidence. The case (unit of analysis) here was the child affected by a life limiting condition and their family. Grounded theory (Glaser and Strauss, 1995) provided the rationale for choice of cases (sampling), data (information collected because of its relevance to the research question), data gathering processes and analysis techniques.

Essentially data were collected through topic-guided interviews, participant observation and participants (particularly the children and their siblings) were invited to use their own artwork, photographs or video (either from the family collection or specifically taken for the research) to help them explain about their day-to-day experiences. The objective was to encourage the children to find ways to represent themselves and their experiences, whilst using the research role to keep participants focused on the subject under investigation.

Chapter four introduces the reader to the life stories of life limited children and their families through a series of five conceptual cases and begins to reveal that, although being affected by a life limiting illness is not one experience, there were common recurring patterns within the life stories of participating children.

In chapter five the theoretical concept I have called 'moments of realisation' is introduced and discussed in detail. Concepts of trajectory in relation to the life stories of children taking part in this research are also explored. The chapter illustrates how some aspects of the theoretical concept of trajectory might be helpful to understand the course of life limiting illness in childhood but also explores some of the problematic aspects of the somewhat linear nature of the concept of trajectory in comparison to the complexity of the course of life limiting childhood illness.

Chapter six examines communication and awareness amongst the children and their families. It reveals how the subject of the child's life limiting illness was managed in smaller less direct topics (probably unconsciously) by the children and families, the role of personal and shared experiences for life limited children and their families, and how participants described the wider context of the lives.

Chapter seven focuses on the voices and experiences of the children. It explores the idea of the child's developing sense of self and how the illness was constructed and explained by the children and families. In this chapter I also explore how families worked to know and represent the children who were less able to represent themselves.

Chapter eight draws together ideas from the findings of the study especially for facilitating talk between children and families when the child is affected by a life limiting illness.

Chapter Two

2 Literature review

2.1 Search strategy and challenges

The purpose of this review was to highlight what is known about the needs and experiences of life limited children and their families especially from the child's point of view. Literature about children with limited life expectancy and their families is scant and fragmented (Cooley et al, 2000; Corr and Corr, 1988; Gold, 1997; Sheldon and Speck, 2002). The range and variation even within a single diagnosis means that using diagnosis as a category for searching the literature only identifies information about selected groups not the wider population of life limited children and their families. In addition, children who are affected by conditions that limit their life expectancy have a range of physical, developmental and emotional needs and the multi-agency, multi-disciplinary nature of support required further complicates the task of finding relevant studies.

2.3 Searches and search terms

Searches included available literature from about 1960 to 2007, using the bibliographic databases listed below. Reference lists from key papers were examined for further sources to follow-up. Discussion with professionals and colleagues generated additional sources of information, some of it 'grey literature' including unpublished works. Databases searched included: Southampton University Web Cat; Bids IBSS; Medline and Cinahl. The same terms were applied to the Department of Health POINT Site and internet sites for key journals including: Death Studies; Children and Society; Disability and Society.

The following search terms were used and combined with the words child, children, adolescence and paediatrics (or 'pediatrics'): life limiting condition; life threatening illness; end of life; dying; terminal illness; chronic life threatening illness; neurological illness; non-malignant condition; limited life expectancy; death; degenerative illness; disability; cancer; malignancy; oncology; severe disability; child's perspective and cancer; child's perspective and chronic illness; child's perspective; child and research participant; families and qualitative research; palliative care (limited to English language and reviews); hospice and respite.

2.2 Inclusion and exclusion criteria of papers for critical review

The review includes studies concentrating on the experiences of children and families where the children had life limiting conditions. Studies focusing on the experiences of dying children or family memories of caring for a child who died were also included. A table of research evidence is appended and includes all the studies identified by the literature search in alphabetical order by the surname of the first author. Selected studies are reviewed. A separate table of key discussion papers is also appended. This table includes discussion from the literature about involving vulnerable children in research and debate about the issue of talking to children about death. This table is included because much of the literature related to this study exists in the form of debate rather than research. Selected papers are referred to in the review.

Studies examining the experiences of 'disabled' children or children suffering from 'chronic illness' were excluded if the paper did not indicate any threat to the children's life expectancy. Studies of bereaved families were also excluded where the focus was on the experience of grief, because this research is about living with the threat of death rather than its consequences. Studies investigating the medical records of children who died are included in the Table of Evidence but not in the review. Whilst the information is valuable it does not come from the child and family's perspective. Authors discussing issues of communication with life limited or dying children are cited in the review to illustrate current debates on talking to children about death and summarised in the appended Table of Discussion Papers. This table is included because much of the literature comes from practitioners who have experience of working with life limited and dying children although the papers do not describe original research.

The review begins with an introduction to four developmental theories debating whether and how they apply to research involving vulnerable children. The review then explores; Concepts of dying and illness trajectory; Awareness and communication about dying; What is know about the experiences of dying children and their families; children with malignant illness and their families; and children with life limiting conditions and their families.

2.3 Theoretical and developmental issues

This section describes the theoretical context for involving life limited children in research. The aim is to create a backdrop for the literature review and for the research methodology described in the following chapters. Four theories of child development are focused on because:

- Psychosocial theory (Erikson) focused on personality development through a series of eight crises and has influenced much thinking about how identity emerges, especially during adolescence
- Cognitive development theory (Piaget) has influenced the development of health, social and educational systems of support for children in the United Kingdom and influenced accounts about how children come to understand death and dying
- Socio-cultural theory (Vygotsky) paid attention to the development and experiences of children with complex needs
- Ecological systems theory (Bronfenbrenner) is frequently referred to and used by researchers investigating the experiences of children.

2.3.1 Personality and identity development

The psychoanalysts put forward a series of possible signposts to help us understand more about the general path of human personality development. Whilst these theories highlight milestones Thomas (1992) argued that they failed to reflect personality development in all its variety and complexity. Freud has been criticized for his emphasis on the role of sexual desires and libido, for his focus on the male experience and for his lack of attention to the separate and different experience of females (Thomas 1992; Berk, 2002).

Accepting the criticism that much of Freud's Theory was based on adults with neurosis Erik Erikson sought to understand more about the development of healthy personality. Unlike Freud, Erikson did study children directly, those of normal development and others described as 'disturbed' (Miller 2002). Erikson described a series of crisis for personality development that he suggested, followed a basic pattern (Thomas 1992).

Although he described these phases as genetically determined Erikson (1963) also highlighted the potential for modification of these eight basic crises through the influence of environmental and cultural factors.

The first crisis, 'trust versus basic mistrust (0-1 year)' - Erikson proposed that experiences related to the dependability of people's behaviour forms the foundation of the child's trust or mistrust in the first year of life, which they build on for the rest of their lives. The second crisis, 'Autonomy versus shame and doubt' (2-3 years) - Erikson's premise was that as the child gained control of the retention and excretion of bodily wastes they also develop a sense of personal power and control, which leads to a sense of autonomy and awareness of personal abilities. The third crisis, 'Initiative versus guilt' (3-6 years) - Erikson described this conflict as the inner struggle between feelings of guilt and initiative or self confidence (Thomas 1992).

The fourth crisis 'Industry versus inferiority' (7-12 years or so) - At this stage the child is said to focus their attention on play with peers. During adolescence young people face physical, social and emotional change. Young people are said to feel uncertain of themselves and the reactions of others. As a defence they can loose their individuality for a time, grouping together and adopting the 'uniform' and 'language' of the group and being intolerant of outsiders. In searching for themselves they can come into conflict with parents, other adults and siblings. Erikson's theory holds that young people who develop through the fifth crisis, 'Identity repudiation versus identity diffusion' (12-18 or so) - come to value and understand their individuality and place in society. Those who do not he suggests continue to show signs of immaturity and intolerance through later life.

Erikson went on to describe psychosocial development through early, middle and later adult life. The sixth crisis facing people in early adult years is willingness and ability to offer sexual and intellectual intimacy to a companion of the opposite sex 'Intimacy and solidarity versus isolation.' People who have failed to develop this ability are described as more likely to retreat into self absorption. The seventh crisis, 'Generativity versus self-absorption' (middle adulthood) relates to adults who devote their attention to meeting the needs of their family. The final or eighth crisis 'Integrity versus despair' (late adulthood), is the stage of life when in Erikson's view the person with healthy personality development can demonstrate a detached concern for life itself, in the face of death and confronted by the end of life.

Taking Erikson's 'Autonomy verses shame and doubt' - we might reflect on the role of feeding difficulties in children born with disabilities or in those who develop eating problems due to illness. What are the consequences for a child who never gains continence or looses this skill? At the stages when children are thought to gain from social contact with peers or develop self-esteem from adult praise what are the consequences of limited opportunities for play, learning and social contact as a result of life limiting conditions? If it is part of 'normal' development that young people turn to the development of intimate and sexual relationships we might consider the extent to which adults communicate with young people who may die in early adult years about their dreams and desires for loving sexual relationships. Does a child suffering from a condition that limits their life expectancy to childhood ever face Erikson's 'Integrity versus despair' crisis and can they emerge with the kind of wisdom and maturity Erikson's model reserves for old age or late adulthood?

Erikson suggested a model for understanding identity development especially during adolescence in healthy children. James (1993) studied the identities and self-concepts of children with a range of illnesses and disabilities and found that accounts could be mapped in two directions: one towards some notion of normality, the other towards concepts of difference. In one set of families James (1993) found that parents and professionals agreed with classifications of the child as different but essentially belonging to the category of 'normal children'. James argued that this group affirmed variety in childhood. James (1993) also revealed competing definitions of the child between parents and professionals where parents rejected professional concepts of the child and worked to form alternative identities on the child's behalf. Similarly Carnevale (2007) found that families of children requiring mechanical ventilation at home, could create a protective capsule at home for the child's identity formation through the control of information to the child and to others about the child.

James (1993) connected these findings to Goffman's (1990) ideas about stigma and the concept that stigma arises from social relationships rather then intrinsic qualities of particular individuals. It was found that the child's self awareness was drawn partly from cultural factors that contributed to his/her recognition of the significance of differences and similarities between themselves and other children. James (1993) suggested that children's experiences of feeling 'different' in a range of social contexts led them to develop and refine their sense of self and other.

2.3.2 Cognitive development, disability and understanding death

Jean Piaget, a Swiss cognitive theorist (1896-1980) paid attention to cognitive development. He developed theories that set out to explain how children come to know what they know. The central theme of Piaget's theory was that adaptation (changes in the person's understanding and ability to apply what they have learned) is based on the achievement of balance and stability (equilibrium) as a consequence of interactions between the organism and environment (Smith, Cowie, and Blades, 1988). This is the process by which the child makes meaning out of objects, beliefs, ideas or events in their world. Piaget's theory suggests that children move through four broad stages of cognitive development each characterized by distinct ways of thinking linked to chronological age. He called these stages sensorimotor, preoperational, concrete operational and formal operational. The stage concept is important because it means that the child's ability to learn and develop is dependent upon and limited by the stage they have reached at that time (Piaget, 1976).

Various researchers have asked whether cognitive development theory has any application for children with complex needs. Lewis (1987) highlighted that much research had focused on children with specific sensory impairments like blindness or deafness to ask about the relationship between particular aspects of the child's cognitive development and absence or impairment of vision or hearing (Detheridge, 2000; Lewis, 1987; Siegler, 1998). Lewis (1987) pointed to the complexity of the 'group' of children suffering from motor disabilities whose abilities and needs can be so varied and individual. Authors indicate that even when children are studied because they share a specific impairment like blindness there can still be significant differences in the ways that they develop (Detheridge, 2000; Lewis, 1987). In discussing Piaget's thesis that the child's actions on their environment lead to development, Lewis (1987) indicated that even with very minimal abilities, restricted environmental and social opportunities cognitive development can still occur, albeit through different routes.

Davis, Watson and Cunningham-Burley (2000) linked Piagetian ideas about the 'natural' development of children with medicalisation of the lives of disabled children,

> *"Both notions pathologize children who do not achieve universal standardized developmental targets and identify disability with impairment. They also show little awareness of the possibility that these concepts are socially and culturally defined. As a consequence, academic research has been preoccupied with differentiating children on the basis of their impairments, 'measuring children's bodies and minds against physical and cognitive norms' and providing advice from a medical and developmental perspective about good practice for working with children with specific impairments." (Davis, Watson, and Cunningham-Burley, 2000 p205)*

Baylies (2002) highlighted how problematic representation of children with severe disabilities and their families is within notions of human development, particularly where the child suffers from cognitive impairment and communication difficulties. Baylies (2002) referred to concepts of what constitutes a good life and ideals of human development, arguing that those who seem to lack a basic level of capability should not be treated as peripheral or unimportant and she goes on to outline how we cannot sidestep the body and ignore what it is or is not capable of – but urges us to strive to ground our notions of capability in terms of humanity.

> *"Conceptual tools that truly take account of impairments, and do not treat them as special cases, abnormalities or exceptions deserving of care, compassion or pity, can broaden our notions of what it is to be human." (Baylies, 2002 p737)*

Classical theories exist about how children come to understand concepts of death and dying in stages. To understand how children develop ideas about death and dying many researchers have utilized Piaget's theories to examine the relationship between the child's stage of cognitive development and concepts of death (Mahon et al, 1999). Classically, authors describe three stages (Carey, 1985; Oltjenbruns, 2001).

> ***The first stage:*** children aged five years and under are said to assimilate the notion of death with the concept of sleep, departure and separation. At this stage

children feel sorrow at the sad departure but do not comprehend the finality of death and separation or the cessation of biological function. They are also thought to have little or no clear insight into the mechanisms by which illness or injury can result in death.

The second stage: children over the age of five and below the age of nine are thought to have a 'transitional' understanding of death. They recognise the finality of death and that once a person has died they no longer exist. They are thought to attribute death to external causes but do not conceptualise the biological consequences.

The third stage: children above the age of nine have been able to describe death in terms of the cessation of biological functions within the body. In reviewing the literature Stuber (1996) argued that age was known to influence children's perceptions of a variety of events and that younger children are likely to be less sophisticated in their concepts of death and dying. However, Matthews (1994) pointed out the majority of children who had taken part in such studies had little direct experience of death. The role of the child's personal experience in forming concepts of death and dying is suggested to be especially important if the group of children to be studied have personal and particular experiences of:

- Their own illness and possible early death
- Death of peers through care settings
- Death within their immediate family unit
- Exposure to war or social violence
- Education and cultural approaches to death

(Mahon et al, 1999)

Judd (1995) suggested that children above the age of six, with actual experience are likely to have a more developed understanding of death for their age than classical accounts would suggest.

2.3.3 Social development and disability

Piaget set out to understand more about child development through observation of the child's activities in isolation. In contrast Vygotsky felt that cognitive function and development could not be understood outside of the social processes on which it is based. Lev Vygotsky (1896-1934), a Russian psychologist who died young, developed Sociocultural Theory. His works were unknown in the west until they began to be translated in the 1960s, long after his death (Smith, Cowie, and Blades, 1988). Sociocultural theory stresses the role of interpersonal processes and the role of society in providing a framework within which the child's construction of meaning develops (Reiber, 1997). Within this framework people who are more knowledgeable initiate children into their social and cultural worlds. In some ways Vygotsky's ideas go further then Piaget's to highlight the active role of the child themselves as well as those around him/her – in that they embrace the child's potential for growth through collaboration with others. Vygostky's theory holds that language begins as a tool for regulating action and communicating needs and becomes a tool for thought.

Vygotsky's emphasis on the role of language as central to development of new skills and ways of thinking raises interesting questions about the development of children who are unable to communicate verbally. He argued for increased understanding of every child's potential for development and need for support through: social enlightenment; increased quantity and quality of interactions and communications with adults; opportunities for relationships with peers, groups as well as individuals; and full use of what Vygotsky called "psychological tools" (Rieber and Carton, 1993). By tools he was said to mean all available systems for communication; in the 1930s this may have included lip reading and Braille. In this the 21st Century we have access to so many other systems of symbols, and electronic means to help accommodate each child's unique needs and ways of communicating (Detheridge, 2000; Gindis, 1995).

Vygosky conceptualised disability as 'primary' or 'secondary' and a consequence of the interplay between the two. He referred to biological or organic impairment as the 'primary defect' and the reactions of society, social restrictions and implications for social interaction for the child as 'secondary defects'. So the child's disability elicits a social reaction to the child, which in turn limits their opportunities for social interaction and impacts on their development, generally in negative rather then positive ways. Vygotsky was critical of the ways in which disabled children are so often referred to in terms of their disabilities or negative qualities rather then their abilities or potential (Rieber and Carton, 1993). In many ways Vygotsky's ideas seem to have been ahead of their time but similar ideas are beginning to re-emerge through other paths such as social constructionism.

2.3.4 Ecological systems theory: research implications

Urie Bronfenbrenner, an American psychologist created an holistic understanding of the contextual influences on child development. Ecological systems theory describes a complex system of relationships through many different environments that affect the child's development (Bronfenbrenner, 1979). The central element - the microsystem - emphasised the patterns of activities, roles and interpersonal relationships experienced by the developing person in a given face-to-face setting; the child's home is one example, their school another. The mesosystem is used to understand the connecting relationships between microsystems. The exosystem refers to social systems in which the child does not normally participate but influence their experiences. Bronfennbrener's hypothesis was that the person's development is profoundly affected by events occurring in settings in which the person is not present. The macrosystem represents the cultural values, laws, customs and resources, which define how the child's needs are prioritised and met at the inner levels. Bronfenbrenner refered to the temporal dimensions of his model as the chronosystem (chrono – refers to chronological influences or changes over time). Ecological systems theory does not view environmental factors as static but ever changing, to be understood in terms of the timings of changes for the child as well as the types of changes that might occur.

Michael et al (1987) argued that ecological systems theory provides a useful framework for research that seeks to understand more about the impact of childhood cancer on the child and their families. This is proposed because 'children do not exist in isolation', also as a means of understanding interactions between the child, family, and other social contexts like school. Michael et al (1987) argued that changes in cancer treatment have increased the number of children suffering from cancer who survive but with cancer as a chronic life threat. Ecological systems theory then provides a possible structure for understanding the complex social consequences of cancer and implications of treatment over a prolonged period of time. However, Mayall (1996 p44) highlighted how Bronfenbrenner's model of layered environments can just become a backdrop to a positivist framework in research rather than 'components of an agency-structure interaction'. Teo (1997) reflecting on Bronfenbrenner's contribution suggested that the interesting questions related to the complexity of representing and elaborating these interconnections between individuals and society. "To understand the subject as a developing subject in a developing society opens new horizons for the work of developmental psychologists." (Teo, 1997) P201. Llewellyn and Hogan (2000) argued,

> *'Most research in physical disability is cross-sectional and the young person is seen to exist in an environment that is a fixed entity, observed only at a single point in time and presumed to remain constant." (Llewellyn and Hogan, 2000 P160)*

Llewellyn and Hogan (2000) suggested that ecological systems theory provides a structure for understanding more about the many and complex factors that influence the development of all children including those with disabilities, by encouraging researchers to explore in more detail the environments and settings in which the child participates, the influence of interactions and relationships within and between these settings and the influence of other settings in which the child does not take part but can still impact on their daily lives.

This section focused on four child development theories (Erikson's, Piaget's, Vygotsky's and Bronfenbrenner's) specifically because they have influenced

a) Ideas about personality and identity development, especially during adolescence
b) Development of systems of support for children and shaped classical accounts about children's understanding of death and dying
c) Included reference to the experiences and development of children with complex needs
d) Design of research involving children to include a wider understanding about the complex environments within which children live.

This introduction to the literature review highlights the opportunity to understand more about children with limited life expectancy by posing a series of questions about the place of such children in dominant theories of developmental psychology. It is argued that as a group they are largely missing or at least their experiences are not accounted for but if included might enhance concepts of 'difference' in humanity. How do these children face questions about interpersonal and sexual relationships? How do they cope with the hurdles of adolescence with the prospect that their own life may be near to an end? As a society do we share these questions with these children or do we leave them to face their questions alone, in the hope that they may be too young to comprehend what is happening to them? What networks of support exist and how do they interact in positive and negative ways to influence the daily lives of these children and their families? How can we recognise the potential of all children and embrace them within our concepts of humanity? What can we learn with them, about them, and about ourselves?

2.4 Concepts of dying and illness trajectory

The concept of 'dying trajectory' emerged from the studies of Barney Glaser and Anselm Strauss (1968) who studied dying adults and children in American hospitals and generated a theory based on the idea that 'certainty' and 'time' yield four types of death expectations:

- Certain death at a known time
- Certain death at an unknown time
- Uncertain death but a known time when the question will be resolved
- Uncertain death and unknown time when the question will be resolved

Certain death at an unknown time was highlighted as especially difficult for all involved because no one could be certain when death would come.

Glaser and Strauss (1965) suggested that dying trajectories have two properties: duration and shape and the way these properties are perceived by all those involved. The following 'critical junctures' of the dying trajectory were identified:

1. The person is defined as dying
2. Medical staff, the family and perhaps the person themselves, make preparation for the person's death
3. At some point death seems inevitable
4. Dying may take hours, days or weeks
5. The last hours of life
6. The 'death watch'
7. Death

The authors argued that when 'critical junctures' occurred as anticipated, everyone was prepared and the infrastructure of support systems worked alongside the dying trajectory (Glaser and Strauss, 1965). However when 'critical junctures' occurred 'off schedule', everyone was caught off guard, so that individuals and systems of support were unprepared. Copp (1999) examined theoretical concepts of illness trajectory in a study that explored the experiences of dying patients and nurses in an adult hospice. A theoretical model based on readiness to die was formed and it was found that participants often referred to the body of the dying person as a separate entity from the self. This study suggested four modes of dying:

1. Person ready, body not ready
2. Person ready, body ready
3. Person not ready, body ready
4. Person not ready, body not ready

It was argued that the body should form part of any theory of dying alongside the perspective of the dying person; consequences of approach to awareness of dying; as well as certainty and time to death (Copp, 1999). Cohen (1995) highlighted the role of uncertainty for families of children diagnosed with life threatening conditions:

> *"There is also support in the data for the emergent thesis that sustained uncertainty is a source of family stress that is perceived by some parents as a greater threat to family life than the diagnosis of a potentially fatal illness." (Cohen, 1995 p47)*

The concept of certain death at an unknown time was applied to children with neurodegenerative life-threatening illness by Steele (2000). Steele (2000) suggested that families caring for life threatened children who took part in her study entered a social process that she called 'navigating uncharted territory'. The model included four dimensions: entering unfamiliar territory, shifting priorities, creating meaning, and holding the fort. Steele (2000) found that families were able to maintain family life through periods of relative stability in the child's state of health but felt as if they were falling down to the next plateau at each moment of crisis, such as any decline in the child's physical health or the need for new equipment.

In terms of illness trajectory The (2002) found, when communicating, doctors and patients tended to maintain control by focusing on short term treatment options, rather than the long term trajectory and ultimate poor prognosis associated with small cell lung cancer. This study indicated that, given the hopeless prospect of lung cancer, medical professionals and patients found control, stability and purpose in the structure of immediate treatment regimes and hope based on short-term improvements as a result of treatment.

2.5 Awareness and communication about dying

Glaser and Strauss (1965) suggested that people interact in various ways to maintain social order when someone is dying. They described four situations between the dying person and 'others' of what they called awareness contexts.

- Closed awareness – the situation where the dying person does not recognise his impending death even though others do.
- Suspicion awareness – the situation where the patient suspects what others know (that he is dying) and seeks to confirm or refute his suspicions
- Mutual pretence awareness – the situation where everyone knows the person is dying, but each pretends that they are not aware in order to protect the other
- Open awareness – the situation where the dying person and others know they are dying and act on this awareness relatively openly.

Closed contexts of awareness were said to complicate care of the dying but were contributed to by various factors including – for example, the fact that many lay people did not recognise signs of impending death and that families tended not to tell the person they were dying. Closed awareness contexts were said to become unstable and a change in any of the contributing factors could lead to a change in context awareness. It was argued that this transition could be managed smoothly and easily or be abrupt and sudden. Suspicion awareness contexts were found to be extremely unstable because the greater the suspicion the greater the efforts required to avoid the person's efforts to 'find out the truth.' Mutual pretence was said to give those involved a sense of relief in some respects, however the stresses and strains of maintaining mutual pretence soon emerged. The context of open awareness was said to create situations where death and dying could be explored by the dying person, family and professionals in relatively open ways. However, open awareness did not eliminate complexity and awareness of their own dying did not necessarily mean that people understood how long they had to live, or that they had any insights into the dying process. The study has been influential and led to a general shift in care settings towards talking about their death with people who are dying.

2.6 Dying children and families

The review only retrieved two studies that explored the experiences of dying children from the child's perspective (Bertoia, 1993; Bluebond-Langner, 1980). Myra Bluebond-Langner (1980) in her seminal work 'The Private Worlds of Dying Children' studied awareness and communication in children dying from leukemia. Specifically investigating how the children came to know they were dying when no-one told them and how they concealed their insights from parents and carers, she argued that,

> *"The children's acquisition of information about their world and their place in it is best understood as a socialization process, and that their decision not to reveal their awareness reflects their knowledge of the order to which they have been socialized, of how it is acceptable to die in this society." (Bluebond-Langner, 1980 pix)*

This ethnographic study was conducted at a large American teaching hospital (Bluebond-Langner, 1980). Thirty-two children, aged three to nine years, were 'major informants of the study' and eighteen were 'primary informants'. The researcher observed and conversed with the children in the paediatric unit exploring their interactions with herself and 'others'. A modified form of play therapy was used, which allowed the children to reflect on their own behaviour without undergoing direct questioning. Bluebond-Langer (1980) suggested that the children she studied gained information about their illness in five stages after diagnosis:

1. "It" is a serious illness
2. Names of drugs and side effects
3. Purposes of treatments and procedures
4. Disease as a series of relapses and remissions (- death)
5. Disease as a series of relapses and remissions (+ death)

It was noted that experience was critical to the children's transition through the stages but that 'stages' were not fixed or age dependent. Experiences arose from symptoms of the disease and opportunities at the treatment centre to assimilate personal experiences with observation of other children with the same disease (Bluebond-Langner, 1980). As the children passed through the five stages of information acquisition it was also found that they passed through five stages of self-concept from being a well child to:

> **Seriously ill** – in which the children explained their new status of 'seriously ill child' at the beginning of every encounter and offered as supporting evidence either stories of changes in the reactions of 'others' or physical evidence on their own bodies like wounds. At later stages this 'exhibition of wounds' was reserved for the first encounter with new people.
>
> **Seriously ill and will get better** – transition to this stage occurred with the experience of remission and a few rapid recoveries from minor symptoms that the children linked to medication. At stage two children and their parents spent time gathering information about drug therapy. The children particularly sought confirmation that the drugs would make them 'better' and their hopes increased with the passage of time.

Always ill and will get better – the children did not pass to stage three until the first relapse. At this point the children were aware that other people, especially parents, reacted as they had at the time of diagnosis. They began to doubt the reliability of information from adults and preferred to seek answers from peers. The children seemed to conclude from a second remission that one could get sick repeatedly but would always recover.

Always ill and will never get better – the feeling that they would get better faded for the children with further relapses, complications from drugs, and life changes such as never being able to go to school.

Dying (terminally ill) – Only children at stage four passed to stage five on hearing that another child with leukemia had died. Children at stage five took up sedentary activities; play and drawing themes frequently related to death and disease. Reflecting on storybooks the children tended to focus on 'death, disease or violence' even when these were not central to the story.

The children discovered their prognosis whether told by adults or not and interpreted their illness differently over time, from acute to fatal. The children used clues from their environment to evaluate their state of health and in the later stages of their illness the children made links between being 'in hospital' and dying. The researcher described how the children revealed their awareness that they were dying. Disclosure was preceded and followed by a period of silence, announced out of the general context of other activities and tended to give the statements a 'dramatic' and 'inappropriate' air, 'conversations that came out of nowhere' (Bluebond-Langner, 1980 p189):

- The children mentioned an individual who had died or someone in danger of dying
- The children attempted to establish the cause of death by asking a question or announcing their own theories and observing the person's reaction
- The children compared the deceased to themselves.

The children were found to be active players in their social worlds, initiating, interpreting and maintaining social order. The children protected others by concealing knowledge of their own dying, in some cases they maintained this pretence until they died. By assuming the role of innocent, vulnerable, unaware child they facilitated the other person's (parent or professional) place in the reciprocal role (care giver, guardian, and protector).

Bluebond-Langner (1980) used an ethnographic design and spent long periods of time with each child at the treatment centre but no time was spent with the children in their own homes or between treatments. It is interesting that the children only conceived of themselves as a dying child towards the end of their illness. The study provides insights into the child's sources of information and active role in disclosing or withholding knowledge of their dying but does not illuminate the process of communication between the child and professionals caring for them (Bluebond-Langner, 1980). Parents and siblings did not participate in the study and therefore the study provides little or no insight into the perspectives of family members. This was a study of children dying from leukemia that was conducted many years ago; treatment patterns and prognosis have changed.

In a case study of a nine-year-old girl dying of leukemia it was found that the child's drawings could be matched to Bluebond-Langner's stages of awareness in dying children (Bertoia, 1993). Parents of the child expressed the idea that their daughter had an intuitive awareness of her own dying that was outside the stages described by Bluebond-Langner (1980). These notions were compared to ideas expressed by Kubler-Ross (1983) that children often have a 'spiritual' awareness of their dying which they might express through three levels of communication:

1. Verbal
2. Symbolic verbal (stories or poems)
3. Non-verbal symbolic language (play or drawings).

Bertoia (1993) argued that the model proposed by Bluebond-Langner did not include the 'inner or spiritual' experience of the child, which it was argued might be preconscious and expressed in the non-verbal symbolic language of children's drawings. However, Spinetta et al (1981) referred to analysis of children's drawings from the psychoanalytic perspective and cautioned against 'over interpretation' of drawings as an expression of 'inner' or 'hidden' feelings.

There was little or no research found on the subject of how to communicate with children about their own death. One survey of 449 parents of children who died of cancer asked whether parents talked about death with their terminally ill child (Kreicbergs et al., 2004). It was found that 27% of the parents reported speaking with their child about death and none expressed any regrets about these conversations. Of the parents who spoke to their child about dying 25% reported using the word 'death'. Parents initiated the conversation when they sensed that their child was aware they were dying, which in most cases occurred in the last month of life. Mothers tended to report spending more time with their sick child than fathers and were more likely to express regret about not talking to their child about death and dying. Although the study findings are valuable, the survey methods used did not allow the researchers to explore how the parents talked about death with their dying children in any detail. How did parents find the moment to raise the subject of the child's dying? What language did they use? How did the children react? These issues were not explored.

Care of a dying five year old boy is described by Faulkner (1997) who argued that terminally ill children are usually aware of their own dying. Guidelines were offered to help adults respond to the 'natural curiosity' of dying children:

1. To be flexible and straight forward
2. Recognise the importance of non-verbal communication
3. Allow communication but don't force it
4. Respond when children try to initiate conversation
5. Be specific and literal in explanations of death
6. Give the child a sense of being loved and missed and a sense of accomplishment in their lives
7. Empower and involve children as much as possible in decisions around their care and dying

There is still a tendency to focus on questions about whether dying children or their siblings can understand death and dying. However there is a lot of debate in the literature based on the experience of people working closely with dying children which suggests, depending on age, stage of development and personal experience, children are likely to be aware of their own dying and can express their level of understanding given support and opportunities (Adams and Deveau, 1995; Bluebond-Langner, 1980; Davies, 2002; Doka, 1995; Hockley, 2000; Judd, 1995; Matthews, 1994). Although dying children have not been well represented in research, available information suggests that experience of the disease, interventions and personal observations (especially of others with the same diagnosis) may be as influential as age and development in forming children's concepts of death and dying.

Other research identified for this review focused on the experiences and perceptions of families of dying children. The 57 bereaved Japanese mothers who took part in open-ended qualitative interviews as part of the study conducted by Saiki-Craighill (2002) talked about finding their own identity through the role of caring for their dying child, and were said to be redefining themselves from the moment of diagnosis. In a much earlier study Samaniego et al (1977) compared the experiences of 21 children with life-threatening illness (seven), children with non-life threatening illness (seven) and healthy controls (seven). It was found that the greater the severity of the child's illness the more the mothers would focus on the child's physical needs and treatment. Participating children with life threatening illness reported wanting more 'affection' but this need was unlikely to be voiced and was therefore missed. In contrast the children with non-life threatening illnesses reported wanting control and independence and this was thought to be a reaction against the extra dependency on parents imposed by their illness. Davies (2005) revealed how mothers needed time, space and privacy with their dying child and with the child's body after death. Theses studies provide some valuable insights into the dying child's personal perspectives and interactions between the child and their mother but no wider picture of the experience for whole families.

Overall, fathers were poorly represented in research but a grounded theory approach was used to study eight bereaved fathers whose children had suffered from cancer (five), spinal muscular atrophy (two) and Tay Sachs disease (one) (Davies et al, 2004). The researchers used the metaphor of 'battling the dragon' to describe the father's experiences; uncertainty, responsibility and daily disruption described as 'living in the dragon's shadow'. It was found that only the fathers whose children had died of cancer described times of 'hope for the child's survival'. In contrast, the fathers whose children suffered from other life limiting conditions knew from the moment of diagnosis that their children would ultimately die in childhood. However, they did hope for alleviation of suffering and increased length of life for their children. This study was important because fathers tended to be poorly represented in research investigating the experiences of life limited children and their families. It would appear that parents experience similar emotions but might react differently when they are caring for a dying child. However, studies that focused on mothers or fathers provide no picture of families' day-to-day experiences or interactions with each other and outsiders.

In a UK based study, While, Citrone and Cornish (1996b) used unstructured interviews to explore the experiences of 44 bereaved families whose children died of non-malignant diseases. Parents were critical of support based only on the child's needs that failed to see the needs of the whole family. The findings of another grounded theory study of eight families caring at home for children with 'prolonged terminal illness' (also described as neurodegenerative life threatening illness) are described in two papers (Steele, 2000 and Steele, 2002). Parents found that professionals sometimes failed to see the 'whole picture' of their life as a family, focusing instead on 'crisis-induced interactions'. Parents questioned advice if it was only based on immediate problems and the child's normal 'state of health' was not considered or where cumulative pressures on the whole family were not taken into account.

Many of the parents gained confidence in their own place as 'experts' in the care of their child and sought to increase their knowledge by accessing information through a range of sources – for example, the internet, international experts on their child's condition, conferences, and other parents with a child who suffered from the same condition (Steele, 2002). Information became a pivotal factor in parents developing confidence and led to more assertive interactions that were not always met with enthusiasm or even co-operation by professionals. Steele (2002) suggested that, despite the family centred care philosophy promoted in many paediatric care settings, all too often clinical practice maintained the imbalance of power between health care providers and those who needed their services. It was argued that families struggled to find help and then to co-ordinate it in a way that left them feeling in control. The study provided information about communication between parents of terminally ill children and professionals, highlighting tensions and challenges on both sides. It would seem that parents want professionals to be aware of the wider picture of their lives in the context of caring for a dying child, not just the child's condition in isolated interactions. Communication within the family and the child's experiences were not described.

2.7 Children with malignant illness and families

It has been suggested that children with cancer experience less anxiety if they are informed about their diagnosis and treatment early. A mixed method approach (described in the Table of Evidence) was used to explore communication in families of 82 children diagnosed with cancer (van Veldhuizen and Last, 1991). Attempts by parents to control the flow of information were often futile and children discovered their diagnosis from peers or siblings. Children who were not informed about their diagnosis often assumed the worst and participating children wanted to control the frequency and content of discussions about the illness with their parents. It was suggested that communication reduced uncertainty and increased the children's sense of control. The children tended to talk in their own time and used 'hints' which parents and carers needed to notice in order to respond.

In one account of practice, Jankovic et al (1994) described how 50 children, aged six to fifteen years, were told they had leukemia without their parents present. The physician used slides and the idea of a garden with flowers and weeds to explain leukemia to the child. The children were then invited to explain their diagnosis to their parents to initiate family discussion. A series of follow up questions by professionals was used to check the children's level of understanding over time (methods and results are described in the Table of Evidence). The authors compared the poor understanding of the youngest participants to stages of cognitive development but suggested that all of the children and families valued the process and doctor's efforts to talk to the children directly about their diagnosis (Jankovic et al, 1994).

Young et al (2003) highlighted the communication needs of thirteen young people (aged 8-17 years) suffering from cancer. Like Bluebond-Langner (1980) they found that links between age and preference for communication were not straightforward. Some of the youngest children wanted detailed information about their condition and treatment whilst older participants wanted only 'basic' or essential information. The study raised questions about the social position of children and participants themselves described complex issues about the importance of age, maturity, dependency, vulnerability and competence in relation to communication. For example, parents took on executive roles to manage what their children were told, which both facilitated and limited communication with the young people themselves, Young et al (2003) highlighted how,

> *"The young people saw themselves as occupying a marginal position in consultations, and some thought that their priorities were of little interest to medicine." (Young et al, 2003 p3)*

The study highlighted how young people are at times afforded little or no voice in health care settings and illustrated that not only do they (the young people themselves) understand their social position as 'different' to that of adults, they actively manage the impact of this on their own needs. For example, the young people participating in the study of Young et al (2003) were aware that they were not given the same information offered to their parents and dealt with this by accepting it as a shield from difficult information, or by seeking answers from carefully selected individuals who they felt would tell them the 'truth'.

In the context of increased optimism for treatment from childhood cancer accompanied by prolonged uncertainty, researchers have investigated how children respond to undergoing (Stewart, 2003) and completing cancer treatment (Haase and Rostad, 1994) - these studies are described in detail in the Table of Evidence. Haase and Rostad (1994) found that completion of cancer treatment had two faces: 'hope with celebration' and 'uncertainty with fear'. The children's uncertainty was linked to their experiences, the 'cyclical' nature of treatment and fears that the disease would recur. Stewart (2003) found that the children very quickly gained familiarity with cancer treatment and represented themselves as 'seasoned veterans'. Although the children participating in the study of Stewart (2003) could describe many unpredictable aspects of suffering from cancer and the process of undergoing treatment, they focused on the ordinary and routine of their lives. Bossert and Martinson (1990) studied children with cancer and suggested that changes in treatment meant that children and families faced years of uncertainty about on-going treatment and the final outcome.

Children's perceptions of various aspects of health care provision were investigated in a study undertaken by Horstman and Bradding (2002). This study found that the children with cancer wanted professionals to talk to them directly about their diagnosis, treatment and prognosis and highlighted humour as an essential skill for professionals communicating with children. In common with other studies Haase and Rostad (1994) and Stewart (2003) interviewed the children once. Studies based on one meeting between the children and researcher raise questions about whether repeated contact would have influenced the results. Beresford (1997) discussed the 'nature of the research encounter' and described how some children are more relaxed with a researcher they meet more then once. It is suggested that, even when research only allows for one meeting with the children who participate, the researcher should take part of the beginning of the session to build rapport with the children (Beresford, 1997).

Education plays a central role in the lives of most children and time at school can be disrupted by illness. A three year study was conducted to investigate how children with cancer function in school (Deasy-Spinetta,1981). Teachers reported on behavioural questionnaires that children with cancer tended to struggle academically and were absent more often then other children, due to their illness and treatment (Deasy-Spinetta, 1981). The teachers also expressed ideas that children with cancer did not articulate negative or positive emotions and would 'retain a self-protective posture' in interactions. Based on the reports of teachers this study provides no information from the point of view of children with cancer about their experiences of school. In a quantitative study the self-esteem of 13 school age children with leukemia was assessed and compared with the self-esteem of 50 school age children in a control group (Mullis, Mullis, and Kerchoff, 1992). The children with leukemia only had lower self-esteem scores than the control group in terms of their school and academic performances. The researchers argued therefore that children with malignant illness need transitional educational support following their diagnosis and treatment. Focusing on cancer treatment and school performance these studies provide information about single aspects of life for children with cancer however they fail to illustrate the child's array of daily experiences in the context of family life. Use of psychometric measures (Mullis et al, 1992) or proxy informants (Deasy-Spinetta, 1981) did not allow the researchers to hear from the children in their own words.

Research exploring the experiences of families caring for children with malignant conditions has used a range of methodologies. In a longitudinal qualitative study Woodgate and Degner (2003 and 2004) set out to understand the experience of childhood cancer from the child and family's perspective. It was found that support from within the family was vital and maintained not only each person's 'sense of spirit' but had a central role in keeping the family unit together. Woodgate and Degner (2004) proposed that interrelationships existed between the child's symptoms of cancer and the 'family's way of being in the world'. The more the child's symptoms intruded on family life the harder it was to maintain daily life and live with the notion of a healthy child.

Biographical disruption is a concept described by Bury (1982) who argued that chronic illness forced affected individuals to confront pain, suffering and death which are normally only considered as vague possibilities or things that affect other people. It was suggested that chronic illness brings the structures and taken for granted knowledge of everyday life into doubt, leading to a fundamental re-examination of the person's life and beliefs about themselves. Carricaburu and Pierret (1995) found that HIV positive men reconstructed their individual and collective identities incorporating past identities and providing reinforcement of biography. Levealahti et al (2006) studied people with inoperable lung cancer and suggested that alongside biographical disruption there was an alternative or parallel process, which they called 'biographical continuity' allowing for integration of various aspects of the individuals past and present lives. They argued for consideration of disruption and continuity to be considered on a continuum rather then as separate entities.

Mothers participating in the study of Young et al (2002) were said to experience biographical disruption as soon as they noticed something was wrong with their child and this increased with the medical diagnosis. The child's illness impacted on the mother's sense of self, social identity and life roles. The threat to their child's life put everyday roles and concerns under a new light and brought with it for the mothers what the authors called an 'obligation of proximity' to the child. Social depictions of children with cancer may not match with families' experiences. Accounts of childhood cancer in newspapers were compared with the accounts of 20 mothers and three fathers of children with cancer in the study of Dixon-Woods, Seale, Young, Findlay, and Heney (2003). It was found that newspaper accounts tended to eulogize children with cancer; constructing them as courageous, stoical and inspirational. In the same reports parents were described as 'battling the disease'. However, parents of children with cancer reported that their children were not always uncomplaining or cheerful. The parents who took part in this study generally felt unable to voice their situation or give real accounts of their child's reaction to their cancer and experienced role strain as a result.

Siblings of children with cancer have been found to experience behaviour problems. In a quantitative study Barrera, Fleming and Khan (2004) found that 47 siblings of children with cancer experiencing 'behaviour problems' reported fewer difficulties when they perceived that they had good access to social support. In addition Kroll and Jacobs (1995) found a negative correlation between self-esteem and duration of illness, or drawn out deaths for siblings. Siblings participating in the study of Rollins (1990) used drawing and talking to express concerns related to their brother's or sister's cancer and treatment but they also took the opportunity to highlight other stresses within the family. Siblings of children suffering from cancer experienced social isolation and complained that parents were 'over protective' of ill siblings but had fears about confronting negative feelings within the family (Cairns et al, 1979; Rollins, 1990). Open communication, involvement with caring and mourning are said to have positive effects on the self-esteem of siblings (Barnard et al, 2003; Kroll and Jacobs, 1995) but will not always reduce the risks of behaviour disturbance, particularly with younger children who may have no other way to communicate their distress effectively (Kroll and Jacobs, 1995). Siblings of children with malignant illnesses have needs that should be comprehended so that understanding can shape support. Hearing the stories of siblings alongside the voices of other family members might reveal more about the impact of childhood cancer on whole families.

2.8 Children with life limiting conditions and families

The needs and experiences of children suffering from life limiting conditions were the subjects of seven studies, many of the studies focused on groups of children with the same diagnosis. In a phenomenological study of nine children and young people (aged 7-15) who had undergone liver transplant, Wise (2002) asked, 'What meaning do children give to this extraordinary experience?' And 'What meanings do children discover in their everyday lives after transplantation?' Wise (2002) described how young participants focused on the reactions of others to physical signs of their illness, especially peers and Wise (2002) connected this with notions of the 'sick body' upstaging the developing self. The participants were articulate and understanding about the misunderstandings of others, stating for example that peers could not be expected to understand about the experience of liver transplant because they would not understand either if 'it hadn't happened' to them. The young people described their distress at being close to other very ill children whilst staying in hospital. The third theme included descriptions of surgical pain and pain from routine recurring procedures like blood tests and the young people made distinctions themselves, describing how they had to get used to certain things because they would face them all their lives. Like Bluebond-Langner (1980), Wise (2002) found that children consciously took on protective roles for others not sharing every detail of their illness with parents. They described themselves as healthy and normal, recognising the potential barriers to concepts of normal - for example, physical signs of their illness or surgery but balancing these real or perceived obstacles with everyday life, including school, social life and relationships with peers.

> *"Concentrating their energy on their lives as normal children allows them to minimise problems and live life to the fullest. The recipients learn to reconcile aspects of their lives outside mainstream childhood activities and discover a new reality from these divergent worlds." (Wise, 2002 p87)*

Psychological adaptation of children who had undergone liver transplant was assessed in a case control study (Walker et al, 1999). The participants included: 18 children post liver transplant, 18 children with asthma (described as a chronic illness), and 18 children who had undergone non-life threatening surgery. The acute threat to life from a liver transplant contributed to development of post traumatic stress symptoms in the severe range in 11% of the focus group, in the moderate range in 11%, and in the mild range in 56%. It was suggested that the combination of acute life threat (from liver transplantation) and chronic life threat (from the child's underlying disease) lead to more symptoms of post traumatic stress than if children were exposed to one or other.

Researchers have examined how children can be knowledgeable and accepting of the consequences of chronic life threatening illness. Earle (2006) found that children requiring mechanical ventilation at night could engage in everyday activities and compartmentalise the technology as something not central to their lives. The beliefs and attitudes of ten asthmatic children (aged 9-12 years) about management of their illness were studied using grounded theory (Ireland, 1997). It was found that the children's concerns to establish normality had some positive aspects but also led them to accept sub-optimal control of their illness in certain circumstances. The qualitative approaches used by Wise (2002) and Ireland (1997) revealed that children can describe potential 'barriers to concepts of normal' imposed by life limiting conditions and still focus their efforts on establishing and portraying 'normal' aspects of their lives. Rigorous design is difficult to assess in qualitative research. One consideration is the extent to which the researcher provides explicit detail about methodology and strategies used, so that in a broad sense the methodology could be reproduced. Ireland (1997) and Wise (2003) provided detailed accounts of their methods of working with the children who participated in each study. Ireland (1997) discussed how internal consistency within data collected for the study was examined and refers to 'authenticity' whilst Wise (2002) used the term 'credibility'. Techniques included using questions to encourage the children to talk in detail and discussing the interview after it was over, giving the children opportunities to question or confirm what was said (seeking a form of respondent validation).

The psychological functioning, cognitive and social development of children affected by life limiting conditions has been studied through quantitative methods. Straker and Kuttner (1980) compared psychological functioning in ten adolescents (aged 12-16 years) who had cystic fibrosis with a matched control group. The results revealed no significant differences between the two groups. Another quantitative study evaluated the impact of thalassaemia major on the psychological functioning and social behaviour of adolescents

(Zani, Di Palma, and Vullo, 1995). The results demonstrated normal psychological and social development in the study participants and the young people with thalassaemia major did better then their peers in scores for self-esteem, self description and functional coping strategies. The cognitive and psychosocial outcome of 16 children on long-term peritoneal dialysis was investigated in a quantitative study using validated and standardised psychometric assessments Madden et al (2003). The results demonstrated that 67% of the children's scores fell within the average range but 50% of their psychosocial adjustment scores fell within the borderline for abnormal. However, the authors argued the results revealed that children with end stage renal failure experienced better developmental outcomes than had been the case in the past (Madden et al, 2003). In a study using Q method (described in the Table of Evidence) of 35 adolescents with end stage renal disease, Snethen et al (2001) described four themes illustrating the children's perceptions of their illness experience: 1 normalisation, 2 illness intrusion and barrier to normalcy, 3 illness management parent focused, and 4 illness management self focused. Like Madden et al (2003), Snethen, Broome, Bartels and Warady (2001) argued that young people had more positive views of living with end stage renal disease then previously described in the literature.

The studies reviewed indicate that life limiting conditions do not necessarily imply negative psychosocial outcomes for affected children. The quantitative methods used by Madden et al (2003); Snethen, Broome, Bartels and Warady (2001); Zani, Di Palma, and Vullo (1995); Straker and Kuttner (1980) and Walker et al (1999) were previously tested through other research adding validity, standardization and generalisability to the results. However, the measures used did not allow for open-ended interview with the young participants and therefore these studies did not illustrate the young people's personal experiences of living day-by-day with conditions that limited their life expectancy.

Research suggests that hearing their child has a life limiting medical diagnosis is a critical moment even when parents already have fears. Cohen (1995) examined pre-diagnostic reactions of parents to cues that their child was ill until the point of medical diagnosis using a grounded theory approach. Initially it was found that parents reacting to subtle symptoms found explanations grounded in 'normal parenting' experiences. When the child's symptoms could no longer be explained in 'lay terms' the parents sought medical help. Cohen (1995) discovered that the period between parents seeking medical help and getting a diagnosis for their child varied from a few days to three years. It was suggested that for those parents who had a prolonged wait the 'diagnosis' brought with it a sense of relief, even though the news was bad, because it brought some degree of 'certainty' (Cohen, 1995; Woolley et al, 1989). In their discussion of the emotional consequences for families when children are diagnosed with serious medical conditions, Freemand, Copley and Bodensteiner (1987) suggested that 'medical diagnosis' is a point of impact for families. Even if they already had fears, it is still an emotional shock. Parents expressed a range of emotional reactions including shock, guilt especially if the condition was genetically inherited, and anxiety especially about 'putting on a brave face for the child' (While et al, 1996a).

Discussing the potential emotional impact on families of children with life limiting conditions Stuber (1996) argued that the life changes brought by the disease and treatment can have consequences for the child, individual family members and the family unit. Mediating factors linked to the different responses of individuals and families are described as a) those that come from the person and b) those that are outside of the individual (Stuber, 1996). Internal factors include personal characteristics, like age, and relate to the way that people respond to problematic life events. External factors consist of social support and other socio-environmental factors. Families were said to focus on the present but framed by themes of separation and loss. Sourkes (1987) suggested that for families of children with a life limiting condition 'time can no longer be taken for granted' and is therefore experienced differently. The disease is said to 'shatter the orderly and predictable unfolding of time'. Sourkes (1987) argued that for people of any age life-threatening illness brings feelings of 'loss' from the moment of diagnosis through all stages of the illness to death: loss of control; loss of identity; and loss of relationships.

The impact on parents and siblings of caring for children with cystic fibrosis was investigated by Bluebond-Langner (1996) who argued that the impact of a child's chronic life threatening illness on parents and siblings could only be understood through exploration of 'the fabric of family life'. She found, from the moment of diagnosis families worked to contain the 'intrusion' of the illness into their lives for as long as they could. Parents were said to use strategies as follows:

1. Routinization of cystic fibrosis treatment related tasks
2. Compartmentalization of information about the child's condition
3. Avoidance of reminders of cystic fibrosis and its consequences
4. Redefinition of normal
5. Reassessment of priorities
6. Reconceptualization of the future

Siblings based their reactions on evaluation of the responses of parents and affected children. Siblings and parents used 'the natural history of the child's illness' to locate events in their own lives and retelling of life and everyday events. The study did not include the children affected by cystic fibrosis and therefore no picture of whole family interaction is provided. The 'natural history of the illness' included the idea that 'turning points' in the child's illness were marked by social and emotional changes in the family as well as the child's state of health. Bluebond-Langner (2000) identified the following six periods of the natural history of the child's illness:

1. Time of diagnosis through to the first annual examination
2. Time from the first annual examination to the first major exacerbation requiring admission to hospital
3. Recovery from the first exacerbation to a time when admissions to hospital become more frequent and unpredictable
4. Development and increase in complications to discussion with the physician that the disease had advanced
5. Increased deterioration through to the conference where the physician told the parents that the child was terminally ill
6. Terminal illness through to death

Although these 'periods in the natural history of the child's illness' illustrate the course of cystic fibrosis and the impact on siblings and parents, the stages or phases described are also defined through systems of support – for example, routine and emergency admissions to hospital. This suggests that for families the trajectory of cystic fibrosis was aligned with critical changes in the child's state of health alongside the medical system of response.

A qualitative exploration of the whole families' experiences and support needs when caring for two or more severely disabled children revealed that families wanted professionals to understand what family life was like on a daily basis (Tozer, 1999). Parents were especially critical of professionals who 'just talked' or meetings assessed to be no more than 'talk shops'. In the 24 families studied the children's needs necessitated at least two carers and this was reflected in unemployment of both parents. Parents struggled to protect siblings from the responsibilities of caring but were forced to rely on their help at times (Tozer, 1999).

Repeated studies have suggested that the child's health on one day was less significant then the overall prognosis and illness 'pattern' on parental adjustment (Dolgin et al, 1990; Tozer, 1999 and Mastroyannopoulou et al, 1997) methods and results are described in the Table of evidence. Children participating in the study of Dolgin et al (1990) were grouped as those with chronic life threatening illness, chronic non-life threatening illness, and a healthy control group. Parents of children with chronic life threatening illness were most likely to use management strategies that responded to the child's overall state of health (illness course, prognosis, physical impairment and time since diagnosis) rather then their diagnosis.

Research focusing on the specific experiences of fathers of children suffering from life limiting conditions is very limited. The study of Davies et al (2004) has already been discussed. In a qualitative study McGrath and Huff (2003) used open-ended interviews and thematic analysis to examine the experiences of four fathers whose children were undergoing treatment for leukemia. The fathers participating in the study of McGrath and Huff (2003) expressed painful emotions but indicated that they did not often show their feelings. They felt that their partners gained strength from the belief their husband was 'in control'. The fathers managed their own turmoil by 'taking time out', working harder or crying alone. Like mothers of seriously ill children in other studies, the fathers re-evaluated their life and career goals and expressed special concern about the separation of family members during treatment, particularly for siblings.

Psychological function of ten healthy siblings of children with chronic life threatening illness was investigated by Stewart et al. (1992). The participants were siblings of children using a children's hospice. Methods are described in the Table of Evidence. Participating siblings reported feeling negative about mothers being less involved with social activities and fathers being less involved with the extended family. Six of the ten siblings were very involved with caring for their brother or sister and the study revealed that siblings tended to accept their own roles when their brother of sister had a life threatening illness.

2.9 Summary

Concepts of dying trajectory originate from the work of Glaser and Strauss (1965). Within non-categorical research, concepts of 'trajectory' tend to describe the course of chronic illness in terms of short and long term consequences for sufferers over time. Therefore researchers describing illness trajectory commonly speak about periods of stability or decline as well as referring to certainty and uncertainty about the likely moment of dying. The review highlighted that concepts of illness and dying trajectory have been applied to children affected by life limiting conditions (Steele, 2000; and Bluebond-Langner, 2000). In both studies the accounts of affected children were not included. Copp (1999) argued that the perspective of the 'ill or dying' person should be central to any theoretical model of 'trajectory' and suggested that the person's body is generally overlooked in theoretical concepts of illness trajectory. This is interesting because the child's body is almost always overlooked in theories of child development.

Children with malignant illness (Stewart, 2003) and children suffering from other life limiting conditions (Ireland, 1997; Wise, 2002) have been able to describe many uncertain aspects of their disease and treatment through research. Yet they focus on the 'routine and ordinary' aspects of their lives as 'normal' children and work to establish 'normality (Ireland, 1997; Stewart, 2003; Wise, 2002). These studies were based on one meeting between the children and researcher. It is not clear whether repeated contact would have influenced what the children shared and therefore impacted on the results.

Research suggests that children with life limiting conditions need a range of information (Bluebond-Langner, 1980; Decker, Phillips, and Haase, 2004; Horstman and Bradding,

2002) and want parents and professionals to talk to them (Young et al, 2003). Parents of children with cancer are advised that their children fare best when they are kept informed about their diagnosis, treatment and prognosis (van Veldhuizen and Last, 1991). However, available research provides very little detail about how professionals and parents should communicate with terminally ill children about death. There is virtually no advice about whether or how to talk to a child with limited life expectancy (who may live for many years) about their diagnosis, prognosis and death.

Available information indicates that when children suffer from malignant illness and other life limiting conditions the whole family is affected and interrelationships might exist between the child's illness and family function (Gravelle, 1997; Woodgate and Degner, 2003). Furthermore individual family members experience disruption to their own life stories (Young et al, 2002). Other studies demonstrate that parents of children suffering from malignant illness and other life limiting conditions manage by focusing on the child's general state of health and illness patterns (Dolgin et al, 1990; Mastroyannopoulou et al, 1997; Tozer, 1999) rather than short term fluctuations or the child's diagnosis. This indicates perhaps why parents want professionals to see the wider picture of their family life (Steele, 2002; Tozer, 1999; While, Citrone, and Cornish, 1996a).

The review indicates that caring for a child suffering from a life limiting condition can generate extreme emotions in parents; requires effort upon effort for the whole family to adjust and then readjust to changes in the child's medical condition; has consequences that affect each individual in the family; and that families want professionals to look at a more comprehensive picture of their lives.

Children living with malignant illnesses and their families have participated in research. Although research is argued to be vital for advocacy at all levels, there is limited empirical research about the experiences of children with life limiting conditions and calls for more sophisticated research design. Children suffering from the wide range of medical conditions that can limit life expectancy to childhood, particularly those with complex disabilities and communication difficulties, are barely represented. The voices of life limited children and their families about day-to-day experiences are largely missing.

In the context of changing patterns of childhood deaths and survival from once fatal medical conditions, many children face prolonged uncertainty combined with profound needs. Straker and Kuttner (1980) highlighted that studies of children with life limiting conditions tend to focus on the acute or terminal stage but rarely investigate the child's experience of chronic life threat over long periods of time. The population of life limited children and their families is not clearly defined and their needs and experiences are not well understood. The review did not identify any study of the day-to-day experiences of children who suffer from life limiting conditions and their families from the child and family's perspective.

Chapter Three

3 Methods

3.1 Aims

The purpose of this study was to understand more about the needs and experiences of life-limited children, some of whom were unable to communicate verbally. The aims of the study were stated in the introduction and are restated here for the reader:

1. How do families face the dilemma of whether or how to be open about the life limiting nature of the child's condition within the family?
2. What is the impact of having a life limiting condition on a child's developing sense of self?
3. Do children who are unable to communicate verbally have active and/or passive roles in shaping their identities?
4. What are the current roles of support services like children's hospices? What are the challenges and opportunities for developing support that matches the needs of life limited children and their families more closely?

A user involvement strategy was incorporated into the development of this design. The study was developed in collaboration with two bereaved parents supported by a children's hospice service (not the host children's hospice) over a number of years. These parents reflected on their experiences and said,

> *"Our experience of 'service providers' ... was that they had very little conception of our situation and were governed not only by statutory infrastructures but by a mind-set that felt that they knew best, and not the parents. They listened but did not truly hear."*

They confirmed the need for and appropriateness of the methods selected for this study.

3.2 Ethical and methodological implications

3.2.1 Stage of development and the role of experience

Staged and continuous approaches to development argue respectively that children develop in quantifiable steps or slowly and continuously over time. Erikson and Piaget emphasised the stages of child development correlated with chronological age, so that one could identify definite or measurable differences between children at different stages of development in chronological age groups. In contrast Vygotsky's theories call attention to more gradual changes over time. Miller (2002) stated that Bandura considered attention to 'stages' as 'counterproductive' because it draws awareness away from individuals with all their differences.

In exploring the idea of continuous change and development throughout life, Sugarman (2000) used the metaphor of a river, 'having a force and momentum of its own but being shaped by the terrain over and through which it flows and the notion that development described in terms of achieving 'norms' carried with it the implication that people develop best when they attain similar 'standards'. Burman (1999) highlighted how developmental theories, which focused on milestones of normal development, tended to marginalize children with complex needs and their families, arguing that they created worry, competition and comparisons amongst parents,

> *'Development thus becomes an obstacle race, a set of hoops to jump through, with cultural kudos accorded to the most advanced, and the real or imagined penalties of professional intervention or stigmatisation if progress is delayed." (Burman, 1999 P58)*

3.2.2 Differences and power: child and adult research participants

The western construction of childhood in which children are defined as 'different', 'other' and 'dependent' (James and Prout, 2001) has led researchers to focus on information from adults about children. Recently children's views have been sought directly. This move was mobilised by the UN Convention on the Rights of the Child, ratified by the British Government in 1991, and in particular Article 12, which refers to the right of the child to express themselves on all matters of concern to them (Alderson 2000). Mayall

(2002) argued that if we are to understand how society functions, children's accounts are as vital as the narratives of adults, because they represent an important social group that exist within different social cultures. Those who study the sociology of childhood take an analytical approach to assumptions about differences between children and adults based on developmental accounts (Holt, 1975; James and Prout, 2001; Mayall, 2002). It is suggested that children might be 'different' to adults but are not lesser persons and that difference does not equate to incompetence (Alderson, 2000; James and Prout, 2001). Greene and Hogan (2005) urged researchers to remain open to understanding how the needs of child and adult research participants are distinct and diverse in some respects and similar in others.

Undoubtedly there are dissimilarities between children and adults, young children and adolescents. Hill (2005) delineated differences of ability and 'power' in terms of social place and influence. It is argued that ability is so varied even amongst children of the same chronological age that it can almost never be assumed for individual children. Much is written about the issues of 'power' and 'status' in social roles. One primary concern about involving children in research is the relatively powerless position of children in society (Alderson, 2004; Holt, 1975; Robinson and Kellet, 2004). Mayall (1996) argued that social order consistently and routinely places children in positions where they are unable to make their own concerns count. Children might not be used to being asked about their experiences or having their views valued. Therefore because of their usual place in society children might try to answer even 'nonsense' questions (Greene and Hill, 2005) be swayed by the opinions of parents or try to please adults. However, as Hill (2005) points out, adults who take part in research can also feel lost or powerless, influenced by how they interpret the status and language of the researcher.

There is a risk of tokenism in research that talks about including children but fails to truly take account of their contributions. Greene and Hill (2005) contended that differences between the abilities of children and adults to participate in research should not be assumed, but they suggested,

> *'The researcher must be open to the use of methods that are suited to children's level of understanding, knowledge, interests and particular location in the social world." (Greene and Hill, 2005 p8)*

There is a sizeable and growing body of literature debating approaches for involving children in research (Coyne, 1998; Fine and Sandstrom, 1988; Greene and Hill, 2005; Grieg and Taylor, 1999; Lewis and Lindsay, 2000; Mohan et al, 1996). Fine and Sandstrom (1988) offer a detailed exploration of different techniques for participant observation by analysis of various projects and through children of different age groups.

The first approach described how researchers tried to become like the children they studied through child-like behaviour and reactions that are more characteristic of children than of adults. Whilst valuing this concept as one worthy of exploration Fine and Standstrom (1988) questioned the ease and ability with which adult researchers could 'become' like the children that they sought to understand. Other researchers have worked alongside children but made every effort to avoid reacting or interacting with the children so that they aim eventually to become almost invisible to the children. This approach was often thwarted by children who were curious about the presence of the adult (researcher) in their midst. The final approach described was one in which researchers aimed to collaborate with the children being studied, to establish a special role with the children, one in which the researcher was accepted or even guided by the children. Researchers using this approach aimed to remain alongside children rather then trying to 'become one of them' but also failed to respond always as an adult might.

In exploring methods that 'work' for children of different ages and abilities, researchers have created methods that include opportunities for children to draw (Arizpe and Styles, 2003; Bertoia, 1993; Bossert and Martinson, 1990; Lange-Kuttner and Thomas, 1995; Rollins, 1990). The draw and write technique was developed by Williams, Wetton, and Moon (1989), to explore the perceptions, explanations and views of primary school children (aged 4-8 years) about what 'makes you healthy and keeps you healthy'. This method has been widely used particularly in health education research (Collins, McWhirter, and Wetton, 1998; Gabhainn and Kelleher, 2002; Horstman and Bradding 2002; McWhirter et al, 2000; Oakley et al, 1995). One valuable aspect of the technique is the standardised instructions, which aim to ensure consistency in the process of data collection. The original study did not seek to analyse the children's drawings. Later researchers have sought methods for analysing the children's drawings at least by cataloguing the items included (Horstman and Bradding, 2002). Backett-Milburn and McKie (1999) were critical of the 'draw and write technique' for potentially failing to appreciate the social context and world of the child and the complex nature of the development of drawing skills in children. However Williams, Wetton and Moon (1989) were explicit that the process was developed for large scale research into younger children's perceptions about a specific topic not their general experiences.

The movement to hear from children about their own experiences raises many interesting questions about how children's voices are used in research (Davis, 1998). Davis (1998) identified three different approaches:

1. To hear from children about 'childhood' assuming one homogenous voice about childhood as a culture different to adulthood.
2. To understand the common meanings and behaviours of groups of children with some shared characteristic - for example, race, replacing illustrations of 'childhood' with representations of the common aspects of groups of children's lives.
3. To understand the complexity of culture and difference, exploring many meanings and voices of children, to understand how they construct relations and have relations constructed for them.

3.2.3 Inclusion of children with communication difficulties

Grieg and Taylor (1999) used the term 'very special children' to refer to children with particular abilities or needs but they argued,

> *'If we accept the special status of all children within society we must also recognise that there are many children who, for a variety of reasons, must be considered to be very special. These children differ from their peers because, for example, they are exceptionally gifted or because of a physical or psychological dysfunction or because they are particularly vulnerable. These children have been, and are, the focus of a great deal of research activity which aims to discover why they are different,*

> *and the effects of their difference in terms of their present and future development. What we should emphasise here is that their rights and our responsibilities as researchers and professionals remain at least the same as for all children." (Grieg and Taylor, 1999 p4)*

Until recently researchers have tended to rely on insights from parents about their children's experiences; this is especially true of disabled children (Robinson and Stalker, 1999; Connors and Stalker, 2003). Ward (1997) concluded that children with complex needs and/or communication difficulties are often not consulted and this is a failing in terms of their fundamental human rights. Children with complex needs and multiple disabilities represent a special challenge for researchers because they may require an innovative and flexible approach to facilitate their inclusion (Lewis and Kellett, 2004; Ward, 1997). On the one hand they are vulnerable and consideration is required for their protection, on the other hand where they have relevant experiences it is important that they have opportunities to take part in research.

Cooper (1999) suggested that, although it is important to understand what it is like to grow up with a long-term illness from the child's point of view, information is limited. Other authors make two important points:

> *'The voice of disabled children themselves is largely absent from disabled childhood research ..." (Robinson and Stalke, 1999 p20)*

> *'It is still rare for research to take a broader approach, asking children about their wider life experiences and aspirations." (Connors and Stalker, 2003 p19)*

In an ethnographic study that set out to understand more about the experiences of children with multiple impairments attending a special school in Davis, Watson, and Cunningham-Burley (2000) worked with children who communicated verbally and non-verbally. The difficulties of working with children who communicate non-verbally are described and the researcher talks of forming a hierarchy in his mind largely based on his own ease of communication with children who could say or sign 'yes' or 'no' to his questions (Davis, Watson, and Cunningham-Burley, 2000). The researcher then sought ways to communicate with the children whose language was more difficult for him to interpret (at least at first) for example, children who used sounds and gestures or other non-verbal cues. In daily contact with the children the researcher learned how to relate with children he had initially doubted were capable of social interaction. The study highlighted that the children who communicated non-verbally were just as likely as other research participants to withhold information until they felt ready to reveal more about themselves (Davis, Watson, and Cunningham-Burley, 2000 p210).

In a study that took a more quantitative approach to understanding the self-perceptions of children with Down's Syndrome, Begley (2000) used pictures and a grid (The Pictorial Scale for Perceived Competence and Acceptance and The School Situations Grid). The researcher chose these tools because they required simple explanation, limited verbal response but relied instead on visual stimuli and manipulation of the pictures. Begley (2000) recognised and tried to reduce the imbalance of power between herself and the children with Down's Syndrome. The children were asked whether they wanted to take part in the study in private (away from their teacher); given time to complete the task at their own pace; and told they could refuse to participate or withdraw at any time. However, Begley (2000) argued that use of 'forced choice instruments' limited insights into the children's self concepts because there was no way to tell whether children chose pictures because they preferred them to the alternative or whether the predetermined categories were perceived by the children as especially relevant to themselves. Davis, Watson and Cunningham-Burley (2000) found that they did not need to create or employ any 'special research tools for communication' with the children. However, time and repeated contact were both vital for the researcher to learn the children's languages (ways of communicating) and for the children to get to know the researcher.

3.2.4 Understanding children in context

To understand the child in a social context the family is central and inextricable, because children experience and participate in the social world, both within and outside their family, which is generally their most enduring social structure (Barnard et al., 2003).

This section can only raise the concept of family in relation to this study, not outline the major theoretical approaches. Family is a surprisingly complex social concept to illustrate, despite the fact that we are all linked to a family in one way or another. White and Klein

(2002) provided a detailed analysis of several dominant theoretical approaches and suggested four typical characteristics of the social group 'family':

- Families tend to have longer lasting connections then other social groups
- Families include people from more than one generation
- Family members are connected by biological, legal and unofficial relationships
- Families consist of small groups of individuals, closely connected and living together and extend to include others associated by 'blood, marriage or adoption'.

These characteristics are no more than helpful indications of the concept of 'family'. In the real world families can include two or single parents, complex structures and 'step' relationships, any number of children as well as, fostered or adopted children. Ultimately families take on many forms but we are all connected to a family by birth, whether the links are sustained or temporary. There are probably timeless connections between ideas of childhood and family (Aries, 1973) and it is a fact of life that we are born helpless and dependent on 'others' for our survival (Holt, 1975). Therefore, it becomes somewhat artificial to separate individuals from the context in which they are embedded for the purposes of research, especially when trying to understand the day-to-day experiences of children.

Symbolic interaction and systems theory suggest that families have rules, goals and roles. If each role in a family has rules (understood by the whole family) then each person has at least some expectations of 'other' family members. Families have access to and use an array of recourses to maintain equilibrium, rules and roles (White and Klein, 2002). Within systems theory 'family' is understood as an organic structure consisting of component parts that operate both independently and as part of the whole. Critical to understanding families in terms of systems theory is the idea that families are separate from their environment, but influence and are influenced by many contexts in which they are embedded (White and Klein 2002). Separateness and boundaries are inter-related. Families tend to live within physical boundaries (the home) and children, by and large, spend much of their lives at home or within other physical boundaries ostensibly for their development, protection or 'well being' (for example, schools, hospitals and hospices).

However, not all boundaries are physical and might be reflected in the ways that families maintain privacy or are open to 'outsiders'.

3.2.5 The question: perceptions or experiences

Finding ways to include child research participants is (like all research) influenced by the research question. Investigating children's perceptions (thoughts and ideas) about a given topic requires a different approach to studies that seek to explore and understand about children's life experiences. Greene and Hill (2005) offered a considered debate about the nature of 'experience' and highlighted that one aspect of experience is the extent to which the subject is conscious of being affected by events. They asked therefore whether preverbal children could be said to have experiences that they are unable to report on (Greene and Hill, 2005) but go on to offer the following thoughts:

> *'Experience is about interpretation, on the part of the self to the self (as in reflexive mental processes) and on the part of the self to others (as in attempts to communicate experience) and, further, on the part of others as they attempt to understand the original experience." (Greene and Hill, 2005 p5)*

This suggests that experience is multifaceted and has some aspects that will be inaccessible to outsiders, either because the subject is unable to articulate their interpretations or chooses not to. An aspect not mentioned by Greene and Hill (2005) is that communication is also multifaceted and might be non-verbal. It would seem that researching the experiences of 'others' is complex and in the end our understandings will be inadequate but one might argue that these issues are common to all research and therefore no reason not to try (Greene and Hill 2005).

3.2.6 Central ethical issues of involving children in research

Authors exploring 'ethics in research involving children' implicitly and explicitly allude to differences between the needs and rights of children and adults implying there are particular ethical issues for research involving children. However, Alderson and Goodey (1996) argued that:

> *"Any extra complications in research with children are common to other 'minority' groups. The main complications do not arise from children's inabilities or misperceptions, but from the positions ascribed to children in late twentieth-century Western societies."*

It is likely that ethical questions emerge from the tension between social desires to protect children and moves in social research to hear from children about their needs and experiences. Morrow and Richards (1996) described the essential dilemma like this:

> *"In everyday social life, we (as adults, parents, or researchers) tend not to be respectful of children's views and opinions, and the challenge is to develop research strategies that are fair and respectful to the subjects of our research." (Morrow and Richards, 1996)*

Alderson (2004) highlighted how standards for involving children as research participants have evolved over the last three decades, in line with changes in national policy. In Alderson's view (2004), researchers who recognise ethical principles as vital components to all aspects of research design are better able to provide flexible methods for listening to the thoughts and experiences of child participants. Whatever the research question, theoretical stance, or methodology adopted, all researchers who include child participants in their studies have to consider the following:

- How they will find and recruit children to the study
- Ways to inform children about the aims of the study and their rights to decline to participate or withdraw at any stage without giving a reason
- Any risks of potential harm or distress to children who take part
- How they will maintain privacy and anonymity for participants.

These questions apply to all research but the practicalities may require special consideration pertaining to the involvement of children.

3.2.6.1 Access

Generally adults like parents/guardians, teachers and social or health care workers are the first point of contact for researchers interested in the experiences of children. 'Adult gatekeepers' are bound to take steps to protect the children they care for. It is common practice to offer adults, with responsibilities for children, information about research aims and plans before seeking their approval to approach children about participation in research (Greene and Hogan, 2005). Alderson (2004) and others have pointed out that adults taking care of children can provide invaluable support to children who do not want to take part in research but might also overpower, silence or exclude children who might otherwise have been willing and capable research participants. In addition, organisations offering support to children and their families are likely to require police checks for all those coming into contact with the children they care for including researchers.

3.2.6.2 Gifts, information and consent

There is some debate in the literature about whether children taking part in research should be rewarded in some way (for example, with money vouchers or gifts). The dilemmas raised by researchers are whether such 'payments' might tempt children to participate in research, when they might otherwise have declined. If young research participants are to be 'paid' for their roles then decisions have to be made about the form of 'payment', as well as the process of giving the reward. Debates exist about whether children should know in advance if they will be rewarded for their role in research.

It is widely recognised that children being invited to take part in research need information about the aims of the study, the process of participation and what taking part will mean for them in practical terms. Increasingly, ethics committees request that separate information leaflets be made available for children being invited to take part in studies - for example, for children in various age groups. All children are different and children in chronological age groups may have different needs in terms of written or spoken explanation. Ultimately, simple, clear written information is more likely to be understood by all participants (children and adults) and can more easily be read and explained to young children or children with learning difficulties (Alderson, 2004).

Children have a right to decline to participate in research or withdraw without giving a reason. In practice, children may not feel able to make these choices or vocalise their unwillingness to participate in research, particularly if parents or other supporting adults are enthusiastic. The issue of children's consent to take part in research is of central ethical concern. The child's age alone is not always a good indicator of competence or likely comprehension of even the most careful explanation. Alderson (2004) advocated an 'opt-in' rather then an 'opt-out' approach and that researchers check with children at various points during the research process that they are still prepared to continue, and be aware that children may reveal their discomfort non-verbally. More recently some authors (Kunin, 1997) have argued that researchers should ask children for their general assent (agreement or willingness) rather than consent (authority or permission that assumes comprehension) to participate in research. Children with cognitive impairment may not be able to comprehend a simple explanation and give their consent or even assent to participate in research. To involve these children, researchers will ultimately be reliant on consent for their participation from parents or main carers. However, children with cognitive impairment may communicate their discomfort with some aspect of the research process non-verbally, relying on family/the researcher to understand and react. Children's consent raises complex questions for which there are no clear-cut answers but awareness of potential difficulties increases opportunities for debate at the level of individual studies and more widely.

3.2.6.3 Possible harm and distress

Although social research of the kind conducted here does not pose any physical or life threatening risks to the children who participate, Alderson (2004) argued that social research could 'worry', 'embarrass', or 'betray' children by fostering false hopes or inaccurate reporting.

There are potential benefits and risks in adopting these approaches. Alderson (2004) described the risks,

> *"The ethical risks of greater participation are that, if children contribute and reveal far more about themselves than they intended, they might later feel greater regret, shame or anger if researchers produce disrespectful reports. Adult researchers still hold the power to interpret and write reports." (Alderson, 2004 p100)*

On the positive side, children who feel that their contribution to research is valued and appreciated, or who experience choice and control in actively participating, are likely to enjoy the process and might gain some sense of achievement that will affirm their self-esteem.

3.2.6.4 Privacy and confidentiality

Hill (2005) identified three elements of privacy and confidentiality in relation to research with children:

1. Public confidentiality – not identifying participants in reports or presentations
2. Social network confidentiality – keeping information the child shares with the researcher private even from other family members
3. Third party breach of privacy – where individuals reveal personal information about other family members. (Hill, 2005 p75)

It is usual practice in research to use pseudonyms for participants. However, some research participants might request to have their own names used. Children in particular might not realise the potential consequences of being named in reports or presentations. As Hill (2005) pointed out, adult views about the best interests of children taking part in research tend to take precedence over the wishes of individuals to be identified by name.

Other data could also lead to identification of participants - for example, drawings, photographs and video; or details very particular to individuals - for example, the disclosure of complex step relationships within families. Hill (2005) recommended that:

> *"Whenever there is doubt, it may be necessary to omit or disguise certain details of a situation so that the persons involved are not identified." (Hill, 2005)*

Researchers sometimes invite children to take photographs or draw pictures that illustrate their experiences or perceptions. Use of image in this way raises many questions about 'privacy' and anonymity. A child's drawing could be recognised by other family members and photographs of individuals are almost certain to reveal the identity of participants to those who know them. In a study of children's perceptions of their home towns, Morrow (2001) decided only to use photographs of places as research data (not people). On the other hand, in exploring disabled children's access to leisure facilities, Murray (2002) argued that 'photography' and 'artwork' provided the best means by which,

> *"the experiences of people unable to articulate with language in any of its many shapes and forms could be directly conveyed to the reader." (Murray, 2002 p7-8)*

In creating her report Murray (2002) decided to include only those images that portrayed study participants in positive ways and restrict analysis to what is described as 'a simple, straightforward level'. Emmison and Smith (2000) discussed the differences between 'artwork' and photography and pointed out that the photograph which is a realistic representation can lead to litigation even from subjects who change their minds at the time of publication. Emmison and Smith (2000) took the position that use of visual data in research rarely depends on readers having opportunities to scrutinise images of the phenomena under investigation for themselves. Emmison and Smith (2000) rely instead on description and carefully selected images. Arizpe and Styles (2003) investigated how children make sense of picture books in which the text or pictures alone would not tell the story and found that children conduct a thorough and detailed visual examination to bring the two things together. Arizpe and Styles (2003) took information about the way that children inspected images and applied the same style of analysis to drawings created by the children participating in the study. Their descriptions are so rich that one hardly needs to see the image as well, although selected drawings are reproduced in the report.

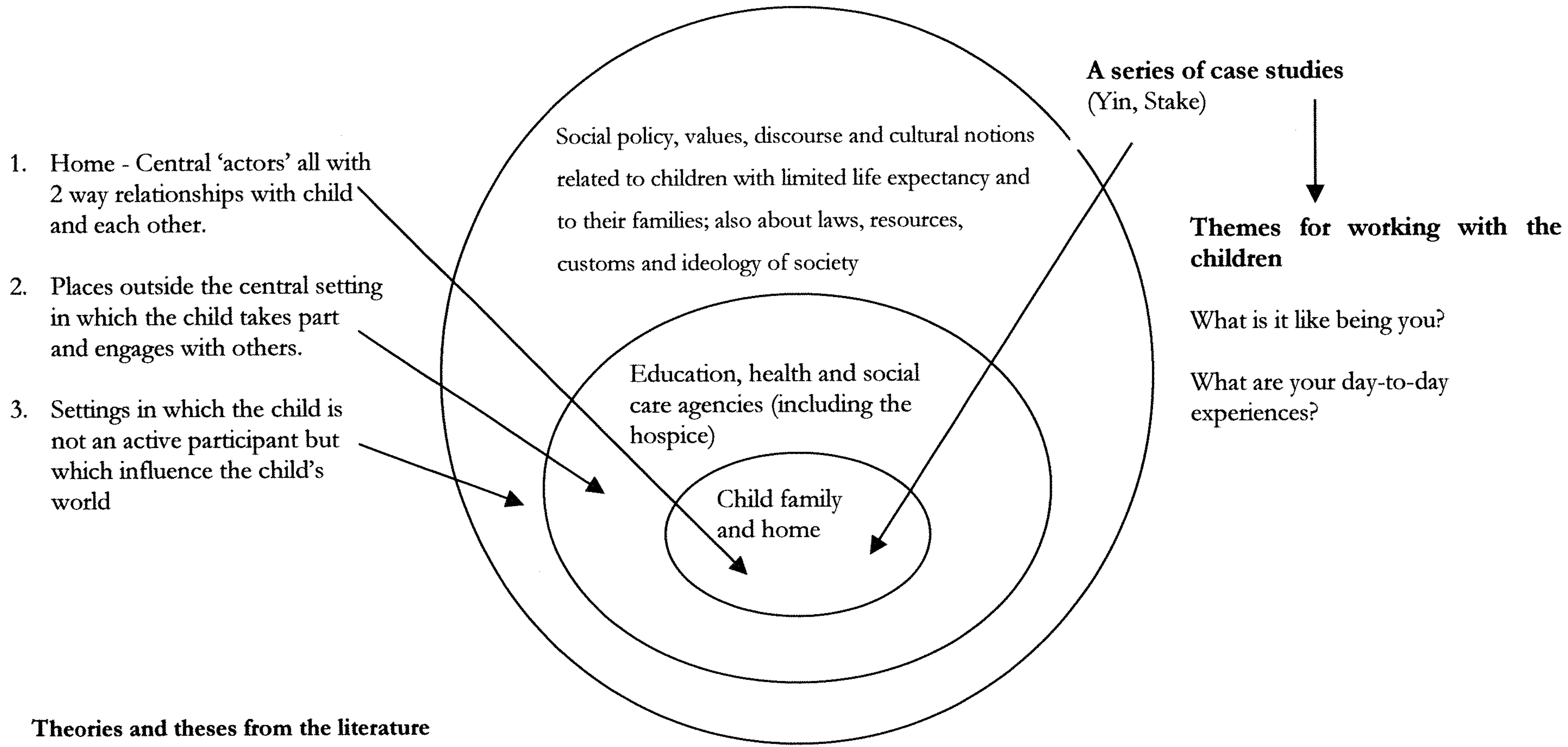
This section has argued that the concepts of childhood and family are so closely aligned that research considering children in the context of their lives should take account of and include links between the child and their family. Theories of family suggest that families have goals, functions, rules and roles and that children actively work within these boundaries to sustain family life and protect family members (White and Klein, 2002). The section has examined the ethical debates, challenges and opportunities for involving children in research, even those with complex disabilities and communication difficulties. It is argued that the challenge for researchers is to find methods and approaches that are flexible enough to include all children where they have experiences that are relevant to the research question (Alderson, 2000; Beresford, 1997; Clark and Moss, 2001; Fine and Sandstrom, 1988; Ward, 1997).

3.3 Design: case study approach

Case study provided the overarching approach for the study and grounded theory shaped the methodology. The research aimed to understand complicated, intricate and sensitive experiences, which for many families extend over years, from the moment of diagnosis to the child's death and beyond it through bereavement. Therefore the study design set out to capture pictures of this experience through a series of case studies, with children of different ages with different prognoses and by looking in detail at the experiences of individual families. The most appropriate number of cases (children and their families) that would offer the widest insight was not clear, due to the complex nature of the subject.

Grounded theory (Glaser and Strauss, 1995) provided the sampling strategy, guided the process of data collection and data analysis scheme. The basic objective of grounded theory is the development of theories from social research (context based field research) that are closely linked to (grounded in) the data, participants and context being studied (Glaser and Strauss, 1995). Ways of working with the children and adults who participated were drawn from classical methods of participant observation combined with interactive methods drawn from education research, especially the Mosaic Approach (Clark and Moss, 2001).

The conceptual framework of the study is illustrated in Figure 1, which describes links between the study design and ecological systems theory, as well as links to key themes drawn from the literature review. Figure 2, provides a visual overview of the design of the research in the form of a 'Mind Map.' This illustrates how the study was framed and bounded. The interpretist and constructionist approach to the study is described in the introduction. With this approach in mind the primary aim of the design was to create a structure for hearing from children and families in their own words, to listen to the experiences and interpretations of participants as well as observing and interpreting interactions between the children and family members.



1. Home - Central 'actors' all with 2 way relationships with child and each other.
2. Places outside the central setting in which the child takes part and engages with others.
3. Settings in which the child is not an active participant but which influence the child's world

A series of case studies
(Yin, Stake)

Themes for working with the children

What is it like being you?

What are your day-to-day experiences?

Theories and theses from the literature

- Dynamic ever-changing nature of the child's environment (Bronfenbrener, 1979)
- Child as capable research participant and wise decision maker (Fine and Sandstrome; Alderson)
- Life limited child as social actor with active roles to play (Bluebond-Langner)
- Construction and reconstruction of childhood (James and Prout)
- Certain death unknown time (Glassier and Strauss; Steele; Bluebond-Langner)

Figure 1: An exploration of the experiences of children suffering from life limiting conditions and their families: conceptual framework

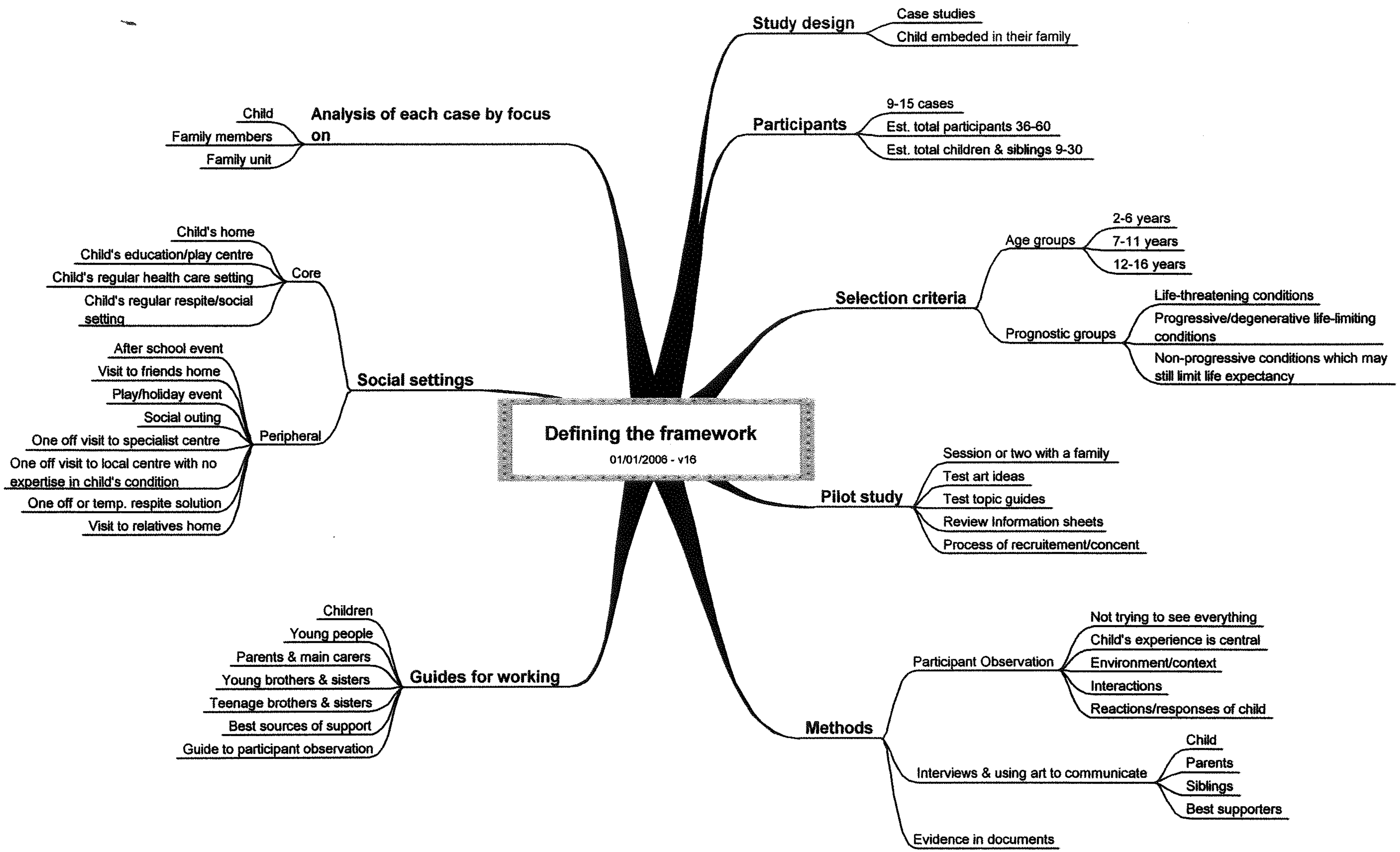


Figure 2: Mind map provides a summary of the study design and describes how this study was framed or bounded

Case study design afforded the opportunity for triangulation (examining the case from a variety of perspectives and sources of evidence). This is reported to have distinct advantages over other methods when phenomena are examined contemporaneously in real life contexts where the researcher has little or no control over events (Yin, 1989). Yin (1989) argued that the first and most important step in designing any study is to identify the type of question being asked. It is argued that case study design is most appropriate to 'How' and 'Why' questions that set out to explore active links over time (Yin, 1989).

3.3.1 Generalisation

Case study research is often criticised for the failure to follow 'scientific' procedures. Positivists might argue that case studies relying on qualitative data and interpretive analysis tend to result in descriptive accounts, which lack calculated assessment and evaluation (Denscombe, 2000). If the case is unique or very particular then evaluation might have limited value. Most widely discussed is the issue of whether information drawn from a single case or only a few cases can have any general application. However, Stake (1995) maintained that valuable insight could come through complete and detailed study of the particular.

> *"It startles us to find our own perplexities in the lives of others."(Stake, 1995 p7)*

Stake (1995) proposed that case studies generate a form of 'naturalistic generalisation' from which the reader gains an understanding of the case specifically because of the detail and depth of description. In this way, case studies allow others to see worlds they are not part of and would never see except through the researcher.

> *'Naturalistic generalizations develop within a person as a product of experience. They derive from a tacit knowledge of how things are, why they are, how people feel about them, and how things are likely to be later or in other places with which this person is familiar. They seldom take the form of predictions but lead regularly to expectations. They guide action, in fact they are inseparable from action. These generalizations may become verbalized, passing from tacit knowledge to prepositional; but they have not yet passed the empirical and logical tests that characterize formal (scholarly, scientific) generalizations." (Stake, 2000 p22)*

3.3.2 Unit of analysis

Much is discussed about the unit of analysis in case study research. According to Yin (1994) the case (unit of analysis) can be an individual, group or community, event or phenomenon. Stake (1995) described how early educational ethnographers first defined the case as a bounded system; an object rather then a process. If we merge the ideas of Yin and Stake we might say that the case is a system of connected parts functioning as a whole (Stake 1995) but where the boundaries between the system and its context are unclear (Yin 1994). In this study the case was defined as a life-limited child embedded within their family. The aim was to understand the perceptions and experiences of the child as an individual, and to frame that understanding with insights from their various family members as well as those of the family as a unit. Each case was unique and represented a different set of social and demographic circumstances contributing to our understanding of the situation in its widest context. The explanations of children and families (where the child has limited life expectancy) about personal, social, historical and political contexts based on day-to-day experiences are at the heart of the research, because experiences from the child and family's point of view were not evident in the existing literature.

3.3.3 One or many cases

Stake's (1995) guidance about selecting cases was to identify one typical or atypical case that could provide information about the system being studied. However there was no way to determine which child and family might represent 'a typical case' or even if the experiences of one child and family would overlap with the experiences of any others without bringing their stories side by side. So a multiple case study design was adopted.

Case study is not necessarily about representation through sampling strategies and it would be irrelevant to seek a sample large enough to inform us about the wider population (Stake, 1995; Yin, 1994). Yin (1994) argued for what he called 'replication logic', suggesting that in multiple case designs each case should be selected because it is predicted to yield similar or contrasting results. Stake, however, maintained,

> *'Balance and variety are important; opportunity to learn is of primary importance." (Stake, 1995 p6)*

3.4 Case selection strategy

The principles of grounded theory were used for sampling, so that it was part of the process of research to recruit new cases until extra data seemed to add no new themes and in general to confirm the on-going analysis. This is what Strauss (1987) called the point of 'theoretical saturation'. Putting balance, variety and opportunities to learn centrally, the selection of cases was therefore based on two characteristics that would be inherent in each and every case: the child's age and the child's prognosis.

Age and prognosis are criteria that appear repeatedly in definitions of life limiting illness in childhood (ACT and RCPCH, 1997; While et al, 1996; and Nash, 1998) and therefore they were primary to the sampling strategy employed. The priority was to try to include three to five children from each of the following groups: young children (aged 2-6 years); children (aged 7-11 years); and adolescents (aged 12-16 years). Within this group of nine to fifteen children, the study aimed to offer equal representation for children whose prognosis could be described as follows:

e) Where cure may be possible but can fail or where the child is susceptible to complications
f) Conditions where life expectancy is anticipated to be shortened (from what is known about the child's diagnosis)
g) Progressive/degenerative disease resulting in shortened life expectancy.

In this way, each case would also take account of at least two significant points of comparison with other similar cases, both within the study, and outside it. The sampling strategy actively sought to accommodate diversity through inclusion of families from different ethnic and socio-economic groups.

It was proposed that informants would be deliberately included to enable exploration of the subject in depth and in its widest context. The most appropriate number of children and families that would offer the widest insight was not clear at the outset, due to the complex nature of the subject, so at various points the data was examined for saturation or repeated themes and the need/feasibility to include further cases was reviewed.

However, an initial sample of nine families was recruited from those supported by the host children's hospice. A minimum of nine cases was required to cover both selection criteria and it was estimated that 15 cases might be the maximum feasible within the resources and time scale of the study (see Table 1).

Table 1 Selection criteria and minimum number of cases required

Prognostic groups	Age groups			
	2-6 years	7-11 years	12-16 years	Total cases by prognostic groups
a) Where cure may be possible but can fail or where the child is susceptible to complications	✓	✓	✓	3
b) Conditions where life expectancy is anticipated to be shortened (from what is known about the child's diagnosis)	✓	✓	✓	3
c) Progressive/degenerative disease resulting in shortened life expectancy	✓	✓	✓	3
Total number of cases by age groups	3	3	3	9
Grand total	9			

3.5 Ways of working

Clark and Moss (2001) offered a framework for 'listening' to the experiences of young children. The 'Mosaic Approach' was used in a study exploring the views of young children about their daily lives in a nursery school: a framework, which they suggested acknowledged the roles of children and adults as 'co-constructors of meaning' (Clark and Moss, 2001).

The mosaic approach as described by Clark and Moss (2001) has two phases. The first involves 'children and adults gathering together documentation' and the second includes 'piecing together information for dialogue, reflection and interpretation'. The mosaic approach describes a series of activities that generate the documents for phase one:

1. Observation of the children by the researcher/key worker

2. Child conferencing or talking to the children about predetermined themes
3. Uses of cameras, mapping and tours by the children to illustrate their perspectives about predetermined themes
4. Interviews with parents
5. Interviews with key workers

These activities generate 'pieces of the mosaic' brought together in phase two for discussion and analysis by the children, parents, key workers and researcher. Talks take place on a one to one basis and in groups. Dynamic aspects of the mosaic approach suggested collaborative ways of working with children participating in research. Most of the children who participated in the study of Clark and Moss (2001) were three or four years old but the same approach was adopted for a toddler by involving her sibling as the photographer. This illustrated the potential for using this framework with children of various ages and abilities and the potential role of siblings as advocates. Drawn from the work of Clark and Moss (2001) was the idea of including various visual tools for the children to represent their own perspectives. The mosaic approach provides a specific framework, within which the children are invited to respond to questions - for example, what are your favourite places in the nursery? However, because my research was about discovering how children and families represent themselves and their own lives in various contexts, it was decided that participants should choose their own ways of working. Anticipating that each child and family (case) would be unique the aim was to know them better, guided by their preferences for styles of working. The ambition was that the study's structure would arise from the 'boundedness' of design and focus on the aims and questions (see Figures 1 and 2). Within this structure, central themes were kept simple, especially to encourage the children to represent themselves in their own ways: 'what is it like being you?' and 'what are your everyday experiences?'

3.5.1 Participant observation

Participant observation originates from the anthropological tradition in which the researcher is part of and participates in the events being studied, witnessing first hand the people, culture and events of interest (Yin, 1994). This investigation used participant observation to embrace the ideas, opinions and perceptions of all the individuals involved; the setting and environment of care; the ethos and rationale related to each event; and outcomes as perceived by the individuals involved. One potential problem with participant observation is the possibility of researcher bias. In this instance I had never played a role in the direct provision of care and was already a daily observer, accepted by staff from the host children's hospice and community team before the study began.

The purpose of participant observation in this study was to be present for periods of time with the child and their family during episodes of their daily lives. I was participating in and part of the events, listening, watching and talking to the people involved. The objective was to take note of the setting, environment, events and interactions with the child and their family, as well as their responses.

Community based observations might be in the family home or other settings - for example, an outpatient appointment or other hospital visit. These would require the co-operation and support of professionals from other agencies, as well as the child and their family. The children and families studied here were participating in many different settings and social situations as part of their daily lives. Holliday (2002) stated, "It is often helpful to think of a social setting as a kind of place." It seemed useful to separate 'core social settings' from 'social situations that may be more peripheral to this study' (see Figure 2: Mind Map framing the study). Children generally spend the majority of their time at home (the place where they live) and it is central to their lives and to their family unit. The child's home therefore was the core social setting for each case. Children have special needs for learning and development and the place where regular play and education takes place was another core setting. Children suffering from life limiting conditions have contact with specialist/experts in health and social care agencies and these people created two more core social settings. Holliday's (2002) reference to 'a kind of place' is useful here because, for some children, education/play, health care and respite or social care might come to them at their own home.

Respite and social care are broad terms used to describe many different forms of support and may in the end have to be defined in relation to each particular case. For example some children attend a regular after school club and they might describe this as a break or respite for themselves. Core settings for the child embedded in their family were identified as listed below:

- Home
- Education/Play centre
- Child's regular hospital/health care setting
- Child's regular respite/social care setting

However, for any child one-off events have a place in their lives and these types of social situations had to be considered on an individual basis. For instance, a child's school play may not seem central to this research question. However, if, for example, the child has for the first time been asked to participate in a school play, the family may feel that this particular event is a defining moment about outsiders' response to their child and it would then become a core social setting in terms of its relevance to a specific case. Social situations that might occur, but may be more peripheral to this study and understanding the day-to-day experiences of life-limited children and their families, included:

- Visits to relatives
- Visits to friends
- One-off or very temporary respite solutions
- After school event
- Play/holiday event
- Social outing

3.5.2 Children and siblings: plans for working

The complexity of the group required methods and a flexible approach that could be adapted to suit the individual needs of all the children, from the youngest, to the most profoundly disabled. To develop the best approach for each child and their family, my skills as researcher were combined with those of people who knew the children well. The aim was to create opportunities to discuss with families - for example, where they would feel most comfortable to be interviewed, whether the children wanted parents or siblings present, as well as details about the process of tape recording particular interviews.

Before meeting the children, I talked to parents about whether/how they explained the life threatening nature of the illness to their affected child and any siblings. This provided me with a baseline understanding about whether or how each family talked about issues of death and dying and shaped work with the children. Personal preferences, attention span and health of each child dictated how I worked with the children and their siblings. The objective was to learn more about the children's ideas, experiences and interpretation of events and interactions in their worlds; not to bring them any agenda through this research but to discover more about the issues that they were already with and chose to share.

In an effort to redress the imbalance of power between myself (the adult researcher) and the children who participated, I invited the children and young people to create artwork based on the topic guide themes. The things that they created were used to facilitate discussion. I prepared some creative ideas - for example, the children might write a poem or make a picture diary. I also collected a series of books to share with the children covering topics like 'my home', 'my family' and 'going to hospital' (an annotated list is appended). I hoped that visual methods would be more like everyday adult-child interactions, and therefore offer the children a chance for control and involvement at a level that was familiar to them determined by their personal preferences.

Before gathering any information from the children with profound disabilities, and especially those who communicated non-verbally, the first step was to learn about their communication styles and skills through observation, especially to understand how others interacted with and understood the child (Detheridge, 2000). Direct communication with those children unable to communicate verbally was through a family member who could help to interpret and confirm whether I had formed an accurate impression.

Inclusion of siblings was central to the holistic approach. Siblings were asked to help me understand about 'being me and having a brother or sister called …' and 'my day or my week'. The siblings who participated in this study were encouraged to interpret these themes for themselves, within their own and their family's existing styles of coping/construction of meaning related to the affected child's diagnosis and prognosis. However, taking part in this research could lead siblings to raise questions about the issues that they face on a daily basis. Therefore I worked closely with the family and their senior care team member from the host children's hospice to support the children and siblings throughout the study and to identify extra support if it was required.

3.5.3 The role of ethics

In June 2002, the proposal was submitted to The South West Surrey Local Research Ethics Committee for consideration. Initially the committee was unable to approve the study at that meeting but raised a series of comments and questions to help begin the process of review and redesign (an outline is appended). In December 2002 I attended a committee meeting of The South West Surrey Local Research Ethics Committee with the Director of Care from the host children's hospice and my academic supervisor. This facilitated further discussion in person and was very constructive from all points of view. Ethics approval was given for this research at the committee's next meeting.

3.6 Pilot study

The research techniques and methods were piloted to see how the procedures for data collection worked in practice; the length of time required; the type and volume of material collected and the feasibility of analysis. Yin (1994) reported that pilot studies are important because they can help to refine the methods and questions and may even provide some conceptual clarification for the research design. Therefore the object of the pilot phase was not to study one or two cases in detail, what Yin (1994) called a 'dress rehearsal', but rather to ask two or three families of life-limited children to help me explore the overall feasibility of the research plans. For example, to review the participant information sheets and interview guides; take part in an interview; agree to a period of participant observation at a venue of their choice; or take part in a general discussion about the plans. The children in these families were invited to help test some of the creative ideas to see whether this was a process that they enjoyed and if it facilitated discussion with me. The pilot study also created opportunities for me to practise general observation of the events and interactions surrounding this work with the children.

3.6.1 Selection and recruitment for the pilot study

Yin (1994) pointed out that participants for the pilot study can be chosen for several reasons unrelated to the final selection criteria. In this instance children and families were identified who might be congenial, accessible and open to the development of a less structured relationship with me (Yin, 1994). They were recruited on this basis.

In July 2003 team leaders from the host children's hospice selected five children and families to approach about participating in this pilot study. Three agreed to participate in the pilot study. The fourth family expressed such a commitment to raising awareness about caring for a child with a life-limiting condition that I invited them to participate in the main study. The fifth family expressed enthusiasm to participate but ultimately school holidays ended, house renovation took over and this family was not able to take part in the pilot. Table 1 is appended and provides a summary of the pilot study, participants, tasks undertaken and lessons learned. Table 2 (Visually linked to the Pilot Study Table: Analysis of Pilot Data, which is appended, by colour coded highlights) illustrates analysis of pilot data related to the conceptual framework of the research design and summarises implications for data collection procedures.

Table 2 Analysis of pilot data

Colour code	Themes	Data collection review of procedures
Yellow	Successes and failures of placing the children's voices at the heart of the pilot study	Work with the child first; negotiate at least some time alone with me for the children; work through a series of sessions, offering different activities (topics) and choice for the child; aim to build enough of a relationship to draw out the child's experiences; don't judge the child's agenda too soon; let the child represent themselves through their artwork
Red	The vital place for parents and their own story	Inherent in existing proposal and confirmed by the pilot study
Green	The vital place of siblings and the value of their contribution	Inherent in existing proposal but aim to value the important role and contribution of siblings with them; let the siblings represent themselves through the artwork
Blue	Children (child and siblings) as capable research participants	Inherent in the existing proposal and confirmed by the pilot study
Pink	General points for the researcher to learn from	A broader repertoire of open questions; be aware of non-verbal communication for all the children.

3.6.2 Implications for conceptual framework and data collection

Pilot study participants shared many perceptions and experiences that would probably be central to the actual research and at times it would have been easy to lose the specific focus of the pilot and begin along the path towards the aims of the main study. However, the purpose was to explore the implications for the conceptual design of the study and proposed data collection plans (Yin, 1994). In many ways this is an artificial separation of issues and procedures that in practice were inextricably woven together. The pilot study confirmed essential aspects of the existing conceptual framework and provided subtle clarifications that are described below. Across the families participating in the pilot study there were common themes, which are discussed.

Overall the 'artwork' helped the children and me to get to know each other and relax in each other's company. No child used the artwork or books enough for me to assess whether they might help children to communicate with me about complex issues. Time was limited and because I only met each child once there was no opportunity to nurture a trusting relationship with me. No child helping with the pilot study shared much in depth, but I was not asking them to. The pilot study did not test the process and methods in this respect but confirmed to me that work with the first family (case) participating in the main study would provide opportunities to explore the depth required to draw out the essence of the child's experience.

3.6.3 The central place of the child

It is complex to keep children at the centre of research, perhaps especially problematic when they participate alongside parents and other family members. I found that parents instinctively adopted a centre stage position and I tended to collaborate with them … so together we shaped the role of the children and siblings who participated in the pilot study. Of these, Fred was only given choices that fitted with the indirect role requested for him by his parents and constructed by myself. In contrast, the parents of Ryan suggested that the voices of the child and his siblings would only be heard through one-to-one interaction with me. Carol was interesting because in some ways her central place was established through the time and close attention required for communication at any level. Nonetheless, it became clear that Carol had experiences and opinions that could be made audible through participation in research. For example, when I asked whether she liked to be at the hospice she said 'yes' looking at me quizzically and when I asked about her favourite thing she said 'Daddy, home' and grinned at me (like many people Carol most enjoys being at home with her family). However, perhaps Martha, Primzanna's mother, highlighted one of the key issues when she said:

> *'Maybe they [children] are not used to being asked but it might be important to try and let them say?''*

It might be useful to add maybe we [social science researchers] are not yet used to asking children about their insights and experiences but it is important we learn how.

All the children who assisted with this pilot study were able to make valuable contributions. They all had relevant experiences and opinions that they began to share.

The children taking part in the pilot study were not restricted so much by their ages, skills, abilities or disabilities as they were by the limitations placed around their roles by adults and by time constraints. I concluded that if children were to have a central role in this research then the design and methods of working would have to emphasize this place.

To achieve this I planned to:

- Begin by offering children and parents an honest explanation of the aims of the research in terms that fit with their current understanding and hopes;
- Increase my repertoire of general questions for the children and consciously try to ask open questions rather then closed ones;
- Review interview proposals so I could give families a clearer idea of how many sessions I would need to have with the children, the length and focus of each session.
- Take more notice of non-verbal communication because it was fundamental to communication with all the children who took part in the pilot study;
- Be aware that every child is different and some may need more time than others to develop a trusting relationship with me;
- Be aware that taking part in this research may uncover things for children and parents that are uncomfortable.

I hoped that subtle adjustment of data collection procedures would reinforce the central place of the children. As a result of the pilot study, I decided that initial discussions with parents should aim to ensure that they understood the aims of the research and likely process, gain consent to approach their children, hear how they explain the child's condition to the child and any siblings, request permission to work with the affected child first and to negotiate at least some time for the child to work alone with me where possible. The aim was to underline the central place of the child from the outset and embed it in the process of working with each family.

3.6.4 The vital place of parents and siblings

In every case, parents began to share information about their child and their own story that underlined how important their place would be in this study. Seven siblings from three families assisted with this pilot study. Although the precise nature of their contribution varied, it was very clear across cases that siblings would be vital to the research. Parents and siblings already had essential roles in the existing proposal and no major review was proposed. However, I was able to value the important contribution of siblings with them, to ensure that they did not feel on the edges of the project.

3.7 The main study

3.7.1 Recruiting families

Care staff from the host children's hospice (referred to as care team members from now on) hold weekly review meetings, attended by a mixture of community and hospice based staff. I attended these meetings on an ad-hoc basis especially to discuss issues of recruitment. Members of the care team identified potential participants.

A member of the care team, well known to the family, made the initial approach to tell them about this research and ask whether they would like to meet me. Initial approaches to families were made during routine home visits or by telephone. No approach was made to children in the family about this project without parental agreement. The same member of the care team introduced me to the family on a joint home visit or during a family visit to the children's hospice. The letter of invitation was given in person (not posted) and discussed at this first meeting. I explained the aims of the research to parents and we discussed their questions and fears, especially those related to letting their children participate. I provided copies of the information sheets designed for themselves and their children, to keep and read through. With their permission I contacted the family in two or three days to find out what they had decided. Some parents declined the three days 'thinking space' and decided at this first meeting that they were happy to participate and gave me permission and written consent to approach their child or children.

3.7.2 Sample

Senior members of the care team were given a blank sampling frame (see table 3 below) and asked to identify children and families who fit the primary selection criteria, of age groups and prognostic categories, in any order.

Table 3 Sampling frame

Prognostic groups	Age groups		
	2-6 years	7-11 years	12-16 years
a) Where cure may be possible but can fail or where the child is susceptible to complications			
b) Conditions where life expectancy is anticipated to be shortened (from what is known about the child's diagnosis)			
c) Progressive/degenerative disease resulting in shortened life expectancy			

Over the first six to eight months of the study a total of nine children in eight families were recruited. All of the families who were approached about the research agreed to participate. Within this initial group of nine, age and prognosis were the primary selection criteria. Secondary aims were to include children who communicated verbally and non-verbally, as well as a cross section of families from different ethnic groups. At that point I asked members of the care team to try to increase the sample to 12 and make it their primary aim to include children who could communicate verbally because they were under represented in the first phase of recruitment. Four more families were identified to approach about the research. One of these families had two children affected by the same life limiting condition. Four of the five potential child participants communicated verbally and the fifth child was learning British Sign Language (BSL). Two families agreed to take part. In the third case the mother said she would discuss the idea with her husband and contact the care team member if they decided to meet me for further information. They did not mention the research again and this was accepted as a negative response. The fourth family was approached on a visit to the hospice. The mother invited me to outline the aims of the research to her teenage son and he expressed interest but had reservations about creating artwork. His mother phoned the next week to say they had decided not to participate. In summary one child who communicated verbally and one child learning BSL were recruited to the study at this final stage in the sampling process.

Overall eleven children affected by life limiting conditions and their families participated in this research. Siblings and parents were essential contributors and there were 39 participants in total. Table 4 describes the number of participants by family membership.

Teenage brothers in two families refused to participate or meet me, an older brother in another family was away at university and two fathers were not available at the time of my visits to the family home because of work commitments.

Table 4 Participating family members

Family members	No participating	Declined to participate	Unavailable at the time of fieldwork
Children	11	0	0
Brothers	3	2	1
Sisters	8	0	0
Mothers	10	0	0
Fathers	7	0	2
Totals	39	2	3

Tables 5 provides the sampling frame completed with details of participating children. All names are false and linked to the conceptual cases presented in chapter four. Individual children are denoted by the use of a letter. These names and identifying letters are used now for consistency and in an effort to obscure real identities.

Table 5 Sample frame showing participating children

Prognostic groups	Age groups			Total
	2-6 years	7-11 years	12-18 years	
a) Where cure may be possible but can fail or where the child is susceptible to complications	Leonora i) Imogen d)	Jagger e)	Marcus j)	4
b) Conditions where life expectancy is anticipated to be shortened (from what is known about the child's diagnosis)	Reuben a)	Reuben b)	Daphne h)	3
c) Progressive/degenerative disease resulting in shortened life expectancy	Jagger f)	Jagger g)	Imogen c) Marcus k)	4
	4	3	4	11

3.7.3 Consent as an on-going process

Participants were identified and recruited by senior members of the care team. In each case it was explained that no child or family had to take part in this study and that they could withdraw at any stage without having to give a reason. Parents were asked for written consent to participate in the study and permission to approach their child/children. Cohen et al (2000) stress the necessity to provide children with a clear explanation of the proposed study and a real and legitimate opportunity to say they do not want to take part.

I asked participating parents how they explained the illness to their child and any siblings, any particular words or language they used with the children and how they coped with day-to-day issues as well as any crisis or changes in the child's health. This understanding helped to frame my work with the children in each individual family. I began (with the first three families recruited to the main study) by keeping initial discussions with the parents to these subjects only, avoiding fuller interviews with them because I wanted to place the children at the forefront of the study even in the process of working. All of the parents responded positively to the notion of putting the children's voices first. This way of working was successful in that it focused attention on the children in each family. However, one risk of working this way was that I could start my discussion with a child from too general a position, failing to draw on their particular experiences and engage their interest and participation. This happened with *Reuben a)* who initially offered short answers to all my best efforts to get into the subject of being in hospital, until his mother began to sit in on our discussions and work alongside me. She had an ability to comment on specific personal experiences for *Reuben a)* who then offered more detail based on what he could remember and I was able to create quiet spaces for us to listen to his reactions. Whilst I remained committed to the principle of hearing from the children first and offering them some time alone with me, I also learned to be more flexible in my approach and I would listen to parents' stories about their child's experiences if they were offered at the first meeting and take particulars with me to the child's interview.

Every effort was made to ensure that the children and their siblings understood the project and their right to refuse/withdraw at any stage. This continuous reflection and review of consent was part of a collaborative approach, which I sought to adopt. I began by explaining the project in simple terms for the young children (aged 4-6). For example, 'Helping with this project means that I will be talking to everyone in your family about what it is like for you and them that you have 'X' (naming their condition, using a family word where necessary). I would like to talk to you first.' For older children and young people (aged 7-18) I began with the words in their information sheet, both naming their condition and referring to it as a condition that 'is difficult to treat or cure'. I offered them the information sheet and we either decided to read it together or I left them to read it in their own time and decide whether to take part or not. The children who communicated non-verbally had a range of cognitive abilities and ways to express their interest, ranging from showing no comprehension or interest to looking intently at the information sheet, having listened to my explanation. For example, the mother of *Imogen d)* gave her a simple signed explanation of the project, I showed her the information sheet and she pointed to the picture of children drawing and signed the question 'Painting about me?'

Ward (1997) suggested it might be helpful for children to practise initiating a break or withdrawal from the study. I used various prompts with each child beginning with the question, 'How will you let me know if you have had enough?' or 'Let's pretend that you don't want to do this project any more.' In practice each child chose a slightly different approach. *Reuben a)* found the tape recorder fascinating and would run off with it at every opportunity. I tried letting *Reuben a)* hear his own voice played back and encouraging him to 'just hold the tape recorder for me' but his fascination with the 'new device' began to be disruptive. As a result in this case I began to be discrete with the tape recorder but occasionally sought consent from *Reuben a)* to continue 'Do you want to stop talking for now?' He seemed very able to make the decision, replying for example, "Yes, let's do stuff now" or "Talk more, I can remember". *Reuben b)* began each session by asking, "Shall I set-up the tape for you?" During the session she would hold the tape recorder keeping her finger on the 'off' button but never electing to press it. *Imogen c)* said, "Well it will sound odd if I start saying I want to stop so I'll just give you the thumbs up sign." We practised this process and *Imogen c)* decided that would be the best way to end the session if she wanted to. Again she never chose to terminate our discussions.

At the start of each session I also rechecked that the children understood the research aims and their right to withdraw. On the whole this was greeted with a giggle or a sigh, 'Oh I know, we did that before - remember!' In practice I found with the children and their siblings that consent to continue was subtle. When the subject of our discussion became intense or difficult for the children, I saw signs of fidgeting, extra swallowing or tears in their eyes. At these moments most of the children created a break for themselves by changing the subject rather then ending our discussion. I might be treated to an anecdote from the day at school or shown a new or favourite toy, but generally the breaks created by the children contained humour or an element of fun. Frequently at these moments the children also moved around physically within the room or in their position. The children initiated these breaks and controlled the length and outcome - for example, they might change the subject momentarily and return to the initial topic, or take a longer break but still return to the initial subject but they rarely terminated the session. I tried to acknowledge their emotion by describing it. For example, 'It looks like this makes you sad, shall we stop?' Sometimes we agreed to work on a piece of artwork at these times. In most cases the children were able to guide me and continue. Some of the siblings had special needs of their own and the brother of *Reuben a)* did not seem able to create these breaks himself, he became quiet and inaudible so I created breaks for him. I learned to keep some 'fun' questions with me for any child who ran into this problem. For example, I would ask 'Supposing you won lots of money what would you spend it on.' The children surprised me in their ability to take the offer of fun and still return to the subject themselves.

Consent to take part and continue was especially complex with the children who communicated non-verbally. For instance *Imogen d)* would ignore my questions when she did not want to discuss a particular subject and was especially skilled at turning things into a joke or game. *Marcus j)* would simply turn his head away from me when he tired of my presence. Three of the children who communicated non-verbally were just as interested in the tape recorder as those who communicated verbally. *Imogen d)* asked about it in sign and her father explained. *Imogen d)* quickly learned to turn it off and would giggle at my feigned distress. *Jagger g)* and *Jagger f)* took the tape recorder whilst I was trying to interview other family members and I left them time to examine it. When *Jagger g)* had finished he dropped it on the floor and wandered off to play. *Jagger f)* handed it back and ignored it after that.

3.7.4 Participant observation

I invited families to suggest 'places' for this process to take place, and they all began with their own homes: 'Well he is here more then anywhere else'. They also suggested other locations for specific reasons: 'We are going on a day out – that might be important for you to see how we go out with all his needs!' Other specific environments included the hospice, and school. The children were not always party to these suggestions but many gained control at the point of meeting. For example, the sister of *Jagger e)* had expressed great interest in participating but sat looking disinterested when I arrived at the family home for our first session together. When I asked whether she had changed her mind about helping with the study she said 'Well no, but I'm too tired to talk much because it's Friday.' We agreed that I should visit on another day of the week and on that occasion I was greeted warmly and we began our discussion with enthusiasm almost instantly.

In all cases I was made welcome and came to feel like an extended family member in many homes. Becoming 'like a friend' and developing a mutual respect with children and families made my role a complex balancing act. On the one hand I was with them to do a 'job,' on the other we were spending time getting fond of each other. I tried to be honest with all the families, especially the children that we might not see each other again after the research process but to honour requests for 'just one more meeting' as far as I could. Whatever my personal feelings, I was aware that I was there to understand the family's experiences and get to know them but I would ultimately be moving on.

None of the families highlighted the number of sessions as a problem. Although in each case parents cancelled visits, all of the families expressed a desire to reschedule my visits and continue with the research process. The minimum number of visits was two and the maximum was eight. In families with more then one or two siblings I found it most effective to work separately with say two children on one visit.

I often found myself sitting on the floor with the children and that just felt like the most natural way to work together. Sessions with the children lasted 20-30 minutes on average and I always sought the child's opinion about 'how many more sessions we needed to have together'. Many of the children gave more detailed information after the first session, some needed only one more meeting with me, whilst others asked for a third session. I realised that children and siblings were taking every opportunity to get to know me and part of that process included observing my interactions with other family members or 'going over' our discussions within the family, 'Oh yes she told us you did that drawing together and she told you about the time she was in hospital for months' *[the mother of Reuben b)]*. Siblings sometimes expressed an openness with the child about their turn to talk to me, 'I told Imogen, Toni will be talking to me and we had a chat about it, she doesn't mind. She said you told her too.' *Imogen c) - sister*

I used a reflective diary to capture my observations about meetings with families. At times I found sessions with children and families difficult because of overlaps in my own life. I found it best to sit down at my own home and hand write notes on my feelings and observations and I typed these in the next day or two. Often in typing field notes I recalled extra details and added them as I worked. Field notes became more than a simple record of observations and events: I used them to explore my own feelings and highlight issues for discussion with my supervisors or counsellor.

3.7.5 Hearing from the children and siblings

I tried to offer each child and their siblings two or three sessions using the appended interview guides to begin and steer each session. I would begin by coming back to key issues from the session before by saying, 'I was thinking about what you told me last time …(giving details).' However, I learned not to over-plan my comments or reactions and began to respond more naturally with each child. I found the children would show signs of surprise or pleasure at these remembered details, sometimes offering more details or just smiling at my feedback. In the process of working with each child I found ways to celebrate their unique or special qualities, with the children where they could obviously comprehend/respond, or with parents for children who seemed less able to comprehend/respond; 'that was very wise that you knew where to go for help'; 'you sound like a good sister/brother or friend to have'; 'she is beautiful, I would like to cuddle her'; 'he is fun to be with'. Where parents wanted feedback on my thoughts about their children I found these useful places to begin, I explained that the detail of children's discussions with me were confidential, which all of the parents accepted, only asking 'do you think they need to talk more' or 'can you help us talk more openly or recommend someone to help us talk more openly as a family'.

I found that the children (unlike the adult participants) were not really waiting to share their story, in fact I think it took some by surprise that I should even be asking. I don't mean that they had nothing to say. In fact once they were sure that I was interested and listening they requested further sessions, in which they concentrated on our discussions rather then any art work. I found the children tended to offer much shorter answers than adults even to open questions. I found four techniques useful in getting the children to expand, I would:

1) Use projection by making the question indirect, for example, 'What do you think this little boy in the story would think of …?'
2) Describe my understanding of their brief reply in two or three sentences and invariably they would have more to say, given my simplistic comprehension;
3) Offer multiple-choice answers to some questions and we discussed their choice of answer together;
4) Leave a brief moment of silence to see if the children had more to add (for the children I adopted the habit of counting to three or four slowly in my mind whilst avoiding eye contact for a moment; in contrast I might count to eight or ten for adults).

Vital to the process of working with all the children was time. We took time to get to know each other and I learned from and about the children over the course of our sessions. In turn the children became more relaxed and open with me over the time we worked together. Much of this developing mutual trust seemed to grow from my presence with the families in their own homes. I observed children watching me interacting with other family members, especially parents. To illustrate, the brother of *Reuben a)*, present but silent when his parents spoke openly about the life limiting condition affecting *Reuben a)*, followed their example by talking candidly at our second meeting. He confided that he listened to 'Mum and Dad talking to me' and felt 'better' about sharing his own feelings. He did not mirror what he had heard but expressed his personal feelings and opinions.

Time with the children who communicated non-verbally and their families was just as essential. In one case I gained the best insights from a visit to the child's school; in a different setting, with other people, I saw *Marcus j)* in a new light. Siblings often acted as advocates. The sister of *Jagger e)* told me that I did not really know her brother and I asked for her assistance. After some consideration she offered to make a video of her brother's day, if I could provide a camcorder and tape. She certainly showed me how much I had failed to see.

All of the children and families took up the idea of using more visual methods for our work together. Each child chose their own ways of working and had preferences for creating artwork by - for example, using photographs or drawings. In the case of *Imogen c)* I assumed because of the level of her visual impairment that she would prefer art materials to feel - for example, modelling clay. I was wrong and she decided to draw. I learned not to make assumptions, just to ask. On the whole I found that simple art materials were sufficient for most of the children and where they wanted more than coloured pencils, paper or the digital camera they asked me 'Can you bring me a blue pen next time? I need it for this writing bit'. Although I talked to the children in terms of us 'working together' on artwork my contributions were only the materials, one or two ideas and encouragement with the process. I did not draw on the child's work, unless they asked me - to for example, 'how do you draw a house?' – which I always responded to on a separate sheet of paper, so they could choose to copy the general idea for themselves. Occasionally I was asked for spellings. We collaborated but I tried never to take over. I made a decision that the children would represent themselves in any way they chose through the artwork, undirected and without unsolicited contributions from siblings, parents or me. For the children who preferred to use photographs, I found it best for us to look at printed copies of their work rather then trying to gather round the laptop screen, with the added temptation of 'playing with the keyboard'!

Drawings and photographs might have been used to illustrate particular concepts or experiences. Inadvertently children illustrating everyday experiences would comment in ways that opened up new subjects. For example, *Reuben b)* announced 'I know I'll draw me in bed, but I'm not sick and I'm not in hospital'. Others seemed to just find it relaxing to 'draw' whilst we talked - 'Oh that's just a pattern' *[Imogen c)]*; 'It's nothing, just a picture' *[Reuben a)]*. Listening to my aims and ideas for working, the mother of *Reuben a)* used family photographs to illustrate various aspects of his life. She provided these as prompts for my work with the children. I went on to include this idea for other families, some of whom used their own family photos in their work with me.

Some children created art before our sessions and then talked to me about their work, whilst others preferred to be creative whilst we talked. Some children used a mixture of these approaches. My role was to keep the children focused on the subject of the research, whatever their creative inclinations. I cannot offer generalisations about creative ideas and for instance children of particular ages. As a group the participating children did not favour one idea above the others - for example, the digital camera was no more or less popular then drawing. The children each had their own particular styles of working and made individual choices.

The children seemed to enjoy the choice of artwork ideas, photography, use of the Dictaphone or reading together, but ultimately I had to offer them a clear and honest explanation of what we were doing together. The creative ideas were fun to some, a sense of achievement to others or just a bore but ultimately the children decided whether to communicate with me or not and I think the primary role of the artwork was to help us 'get to know each other'. Some children used drawings or photos to show what might have been difficult for them to articulate. For example, *Imogen c)* who has learning difficulties took a beautiful series of photographs using the light reflected on her window to illustrate that she is "going to 'Neverland' with Peter". These will be explored in the findings section.

Six of the affected children were unable to create artwork or take part in discussion of choices. I found siblings and others willing to take on this role: for instance a care team member took photographs to illustrate when *Leonora i)* visited the hospice; and *Marcus j)'s* teacher provided copies of his schoolwork (scribed on his behalf by classroom assistants). She also encouraged me to photograph communication aids that worked or did not work for *Marcus j)*. She said,

> *'We tried this one, but it didn't tell us more then he can already let us know with a glance or a facial expression. I expect you need to know that.'*

For children who communicated non-verbally, visual materials gathered by siblings or others tended to trigger further discussions within the family about the child, helping me to know them all better.

In working with the first two or three families I learned to be prepared for the unexpected, because, despite all plans, families can be in chaos, the child or sibling you thought was going to talk to you may be upset for some personal reason or have gone off to play with a friend. On the other hand, the child or sibling who seemed unable to focus on the subject of the research might openup unexpectedly. I have come to think that as adults we are rarely as in control as we assume we are. Even the youngest participants in this study realised what I was focusing on and all the children made decisions about what they would share, when and to what extent. I was never rejected except in the hospice where there were infinitely more interesting things to do and I was told by one child, 'I'm going to find George now because we are going to play on play station but you can come and see me at home, ring my mum OK!'

In addition to choices about artwork, I aimed to give the children and their siblings some control over the location of our interviews. Some older siblings chose to meet me away from the family home - for example, in a local coffee bar or their place of work. The children and younger siblings tended to have less flexibility and on the whole I had to meet them at home, the hospice or their school but this was more often a consequence of where they happened to be on the dates available to us. In these cases I tried to give them a choice within the building, if I could - for example, 'Which room shall we talk in?' Some selected private rooms whilst others chose the family lounge and asked for other family members to leave us to talk, and families were generous in their reactions to these kinds of requests.

3.7.6 Hearing from the parents

The aim was to elicit information from parents about their child's life and day-to-day experiences of the family with the child, using the appended interview guides. Parents were also offered the opportunity to gather and contribute visual material including photographs or their own drawings to illustrate their personal insights and to use these as a trigger for discussion with me. Many mothers listened to this idea and used family photographs to help illustrate family life, including critical moments or changes in the child's condition. Use of existing family photographs in this way became a helpful trigger for discussion. I would work with the individual to understand the meaning and intention from the person's own perspective. It was hoped that this method of contribution might appeal to some of the fathers in particular, who are frequently poorly represented in qualitative studies. In practice the seven fathers who participated in this study all chose to 'just talk'.

3.7.7 People providing support

The initial plan was to invite each family to identify two individuals who provided them with the support which best met their needs, so they could be invited to take part in one semi-structured interview using the relevant topic guide. However, working with the first family to participate in the study illustrated the quantity and quality of information available from the family alone and I decided that to invite comment from people outside the immediate family would add little to the child's story and might only serve to dilute the child's voice. For these reasons I did not approach families for contact with those who provided 'the best sources of support'.

3.7.8 Relevant documents

In each case a variety of different documents could further understanding and be usefully included - for example, case notes held at the host hospice with the permission of parents, correspondence or documents written by children and families themselves. Participants suggested documents and the children and families highlighted different documents as relevant. For example, *Imogen c)* had been featured in the church magazine and she shared this with me, we sat and read it together and discussed the way she was portrayed.

3.7.9 On-going review of data

Within this study I aimed to make review integral to the on-going process of gathering data and confirming its meaning with the subjects of all ages. I worked in partnership with the children, young people and their families. Together we discussed and agreed for example, which creative techniques to use, when tape recording or field notes would be used as a record of each session. Given the slow process of the research and the pressure on families participating in this study, I intend to send families a thank you card when the findings are available, which offers them the chance to send for a simple summary of the findings if they would like to.

3.7.10 Thanking the children and moving on

I decided not to offer remuneration to the children working with me. I simply explained the study as described under 'Consent as an on-going process' and asked if they wanted to participate or not. I wanted their commitment to the aims not to be coloured by any anticipated payment. However, given their enthusiasm, invaluable assistance, input and joyful company, I found it difficult to end my time with families and move on without a backward glance. Therefore, to mark our time together I found a gift for each child and their siblings that I hoped they would enjoy, based on what I had discovered about their interests and hobbies. These gifts were given on my last visit to the family home. It was a pleasure for me to seek out gifts for each child and eased my moving on from families.

3.8 Data management and storage

It is difficult to maintain anonymity for the children and their families. All names have been changed. Throughout the course of data collection I used one set of false names for participants and labelled all data accordingly. At the end of data collection I changed to a second set of false names linked to conceptual cases (presented later) so that real families, raw data and the thesis use different names and I hope it will be difficult to link individuals to the final report. As already mentioned, some families included stepparents or step relationships between siblings and these will not be examined in detail to maintain anonymity. To further obscure the identity of individual families I have added or subtracted one year from the ages of certain siblings. All raw data was stored in a locked metal filing cabinet at the hospice. Only an administrator and myself knew the location of the key, but she did not have any reason to use it. My own code-book of false names and reflective diary are stored separately in a locked drawer.

3.9 Data analysis

3.9.1 Analysis of case study data

Analysis of case study data is inevitably complicated by the range and quantity of information gathered. For this reason Yin (1994) maintained that the case study researcher must have a general analytic strategy:

> *"The ultimate goal is to treat the evidence fairly, to produce compelling analytic conclusions, and to rule out alternative interpretations." (Yin, 1994 p103)*

Yin (1994) described four analytical techniques that he argued were especially relevant to case studies:

- Pattern matching, to compare actual patterns in research data with predicted ones
- Explanation building, to explain a phenomenon and where possible to illustrate 'causal links'
- Time series analysis, looking for patterns in data over time
- Program logic models, to explore the data for 'repeated cause and effect sequences of events' that are somehow connected

Yin (1994) argued that within multiple case study design the researcher should give priority to analysis of each case before conducting cross case analysis. The central question of this research was: what are the day-to-day experiences of children suffering from life limiting conditions and their families? This 'case' was explored with eleven children in ten different families. However, it is important to remember, although real accounts are represented the 'case' is theoretical. The purpose of analysis in this investigation is to find a way to understand and illustrate more about the 'case' but not expose individual families.

In terms of using Yin's (1994) techniques for analysis there were two problems. Firstly data was not collected over time, which ruled out 'time series analysis' and 'program logic models'. Secondly the literature review did not yield any specific patterns to compare the data against, making 'pattern matching' unworkable. The exception is the study of Bluebond-Langer (1980), which portrayed a possible 'pattern' for understanding more about the way that children come to know that they are dying. Whilst this might be very relevant to part of the study, it would fail to illustrate the whole case.

Explanation building is a process that Yin (1994) himself suggested was similar to the hypothesis generating process described by Glaser and Strauss (1995). Glaser and Strauss (1995) provide a more comprehensive description of the process for grounded theory analysis than Yin (1994) provided for explanation analysis. Grounded theory (Glaser and Strauss, 1995) provided the sampling strategy for this research and guided the process of data collection within each case. Therefore grounded theory was also applied to analysis of the research data collected for this study.

The function of multiple case study analysis is to develop a detailed picture of the area of research through focus on each of the cases studied, in turn, before looking across and between cases. Therefore the first stage was to analyse the information about each case through focus on three levels: the child, individual family members and the family as a unit. Data for analysis constituted field notes and other relevant documents; visual materials with related dialogue and interview transcripts. Participants explained visual material and I reflected on individual explanations in light of the collective.

3.9.2 Grounded theory analysis

The goal of grounded theory is to generate hypotheses that account for patterns of behaviour, relevant or problematic to the participants (Glaser and Strauss, 1995). Patterns of behaviour include social processes and interactions. Coding is the process of finding and labelling theoretical or conceptual categories and subcategories in the data. A category becomes a conceptual element of a theory that has properties or elements within it. Categories and properties represent ideas drawn from analysis of the data. Strauss (2003) suggests that researchers should code for:

- Conditions
- Interactions among the actors
- Strategies and tactics
- Consequences.

Core categories are central elements of emerging theories. Core categories account for most of the variation in patterns of behaviour and, once a core category emerges or is suspected, all categories and subcategories are related to the core and each other. Through the coding process the researcher is constantly asking what category, property of a category, or emerging theory does this happening represent? (Strauss, 2003).

The process for collecting and analysing data suggested by Grounded Theory uses the principal of 'theoretical sampling'. Theoretical sampling requires that the researcher collect code and analyse data alongside the process of deciding what and where to collect more data in order to develop the emerging theory (Glaser and Strauss, 1995). Theoretical saturation occurs when further analysis only confirms what is already understood about a category and adds nothing new.

> *"Theoretical sampling means seeking pertinent data to develop your emerging theory. The main purpose of theoretical sampling is to elaborate and refine the categories constituting your theory. You conduct theoretical sampling by sampling to develop the properties of your category or categories until mo new properties emerge." Charmaz, 2006, p96*

3.9.3 Analysis of each case

Data analysis began within each separate case. I used theoretical sampling within each case by comparison over and again of information from individuals and 'within family stories' until no new categories emerged from each participant or the family as a unit. Grounded theory methods were used to identify and index conceptual categories based on phrases or recurring descriptions of incidents or phenomena. The data related to each case was repeatedly re-examined to add as many categories and sub-categories as necessary to reflect all the concepts and relationships within the data as accurately as possible. This large number of categories was refined into core categories for further exploration and theory generation. Triangulation was used to look at patterns across various sources of evidence from several perspectives for breadth and depth of information, in order to build an image that valued all contributions, accepting that children and adults report from their different points of view.

I analysed data from each case using a computer software package, called Atlas.ti version 5. Atlas.ti version 5 was designed specifically for grounded theory analysis and allows for coding of text and images or parts of images. The software does provide a means for condensing codes into themes, which I tried but in the end I found that I think best with paper and my own handwritten notes. However I did use a combination of handwritten notes and the computer software, which helped me catalogue excerpts from the raw data that illustrated the concepts I was developing. I also found the function for memo writing provided within Atlas.ti very helpful for making instant notes about thoughts as I explored data on screen and I used these memos later to expand ideas further.

For each case I ended up with key points about the way that the family members told the child's story and how the child talked about themselves alongside main themes talked about by the whole family.

3.9.4 Analysis of multiple cases

The aim was to achieve cross case analysis (analysis of multiple cases rather then analysis of many interviews on the same subject). Inevitably I took categories and core categories from one case to the next, looking for similar themes, contradictory information and new categories. Analysis went on to look for patterns across and between cases. The aim was to draw on analysis of each case and generate a theory to account for patterns of behaviours across cases.

I took my analysis of each separate case and looked for overlaps with other cases in terms of the way the child's story was explained by the child and family and themes explored by participants. I found that although all the children had different medical diagnoses there were quite a few overlaps in the way the 'trajectory' of the child's illness was described. To provide one example: for some children their deterioration was linked to acute phases of the illness but for others decline was slow and over long periods of time. I took these overlaps in stories and created five conceptual cases based on overlapping aspects of the children's life stories. I also tried to incorporate within conceptual cases some unique aspects perhaps only relevant to one or two participating children. The conceptual cases were therefore the result of cross case analysis and written to introduce the reader to the types of children and families I met and to reveal some of their experiences without exposing the identity of individual children and their families.

3.9.5 Presentation of the findings

The aim of grounded theory is to generate a theory embedded in the study data, which accounts for variations in patterns of behaviour in relation to concepts that are problematic to those studied. When the theory generated is primarily relevant to a specific problem or research question it is said to be substantive. If, however, a theory can be generated which is conceptual and therefore applicable to a wider issue it is said to be formal (Glaser and Strauss, 1995).

The findings are presented as follows: the children and families are introduced through a series of conceptual cases. The case presentations are followed by a chapter that draws out and describes in detail the concept of recurring 'moments of realisation' in the unfolding life stories of life limited children. The next chapter explores how children and families talked about the subject of the child's life limiting illness and the influence of experience and social context on family communication. The following chapter examines the impact of the life limiting illness on the child's developing sense of self and identity as presented by their family.

Although I have included descriptive data about the children's artwork in this report, I have chosen not to reproduce any of the images. When ethics approval was given for the study it was suggested that consent for use of children's artwork should be confined to the specific images to be reproduced in the thesis or oral reports, rather then a general consent to use all of the artwork created for the study for presentations. This could not be done until decisions were made about which artwork was most reflective of the research questions and findings. Over the three year period since the study began, all of the children and families have faced many changes and it seemed inappropriate, given the range of their needs, to approach families facing bereavement and other life changes for a second round of consent to reproduce children's artwork or photographs, especially when I was able to describe the few relevant images in words.

3.10 Summary

This chapter restated the aims of the study and presented a design to meet these aims. Considerations raised by the ethics committee were described and addressed within the design of the study. Detail is provided of the pilot study that clarified the conceptual framework and data collection procedures. Accounts of procedures for the main study are detailed. Specifically I have tried to illustrate 'what I learned from working with the children and their siblings' to make the methods reproducible and to share my insights with other researchers wishing to work with children. I have explained why grounded theory was chosen for data analysis above those methods described for case study analysis and described data analysis within and across cases.

Chapter Four

4 Case presentations

It is impossible to describe the pain and ultimate isolation of families caring for life limited children; in the end they face the loss of their own child in ways that 'outsiders' can never comprehend. All I can do is try to show you some of their practical and emotional day-to-day struggles in the way that these problems were constructed for me by the children, siblings and parents that I worked with. Daily hurdles were innumerable and I witnessed more difficulties than any family articulated.

4.1 The children and their families

I met ten families caring for eleven children affected by a range of life limiting conditions. All of the children had physical and emotional needs beyond the needs of the average child. All of the families were devoted and loving and faced the toll of extra daily demands on each family member and the family unit without complaint and often with a great deal of humour. In all cases family members were protective of the child, emphasising skills and positive personal qualities.

The children suffered from a variety of life limiting conditions and their stories are illustrated through a series of conceptual cases. The children ranged in age from four to eighteen years. There was a mixture of children who communicated verbally and non-verbally in the three age groups (2-6 years, 7-11 years and 12-17 years). Of the children who communicated non-verbally, one was learning British Sign Language (BSL) but had a limited vocabulary, and others used a combination of eye contact, facial expressions, gestures and sounds. For these children I relied on my own observations and the interpretations and stories of family members. A member of staff from the hospice who was fluent in BSL, helped me to communicate with *Imogen d)* supported by her father and mother.

All ten mothers took part in the research. In all cases I had contact with mothers first. Mothers worried about the child and talked openly to me about fears for the child's life but they were at different stages in their experiences with the child and their thoughts reflected this.

> *"But there again I mean I think a few months ago it was mentioned briefly that families would have a memory book if anything happened and I thought like 'gloss over it' obviously we don't need to talk about that now and hopefully never will and I just remember thinking like memory book, I need a memory house." Reuben a) - mother*

They also had concerns for siblings and the whole family. Many mothers were trying to balance meeting the individual needs of all family members and were left feeling torn apart. Mothers also talked about changes in their relationship with their husbands and the near impossibility of spending any time together as a couple. There were seven fathers who took part in the study. Initially many of the fathers took a reserved stance, letting their wives do the talking. However, in all cases they agreed to one-to-one meetings with me and talked at length. They shared their wives' painful emotions and had fears about the child's death. Many fathers who participated also expressed fears for siblings.

There were six brothers in the families who took part in the study. One brother was away at university. In two cases older adolescent brothers refused to participate. One told his mother that the family did not discuss the child's illness with each other and should not be talking to an outsider. The mother acknowledged that this was a true reflection of the family's communication strategy. However, she wanted to know if her son needed to talk but didn't know how to ask him. Three brothers aged ten to twelve took part in the research, two of the three had mild learning difficulties (ADHD and dyslexia). These three boys used initial time with me to talk about themselves and their own experiences but would make statements linking their own experiences to those of their sibling. A brother articulated how his own needs are even less visible than the child's, constructing the idea that a scar would at least demonstrate that *'something was wrong'* to other people.

> *"I'd love to have a scar! Because then people can see what is wrong with you and that can be quite cool" he added "not one like his though that is horrible."*
> *Reuben b) - brother (aged 12)*

All eight sisters from the participating families took part in the research: four were younger aged two, four, eight and thirteen. The other four were in their mid twenties. The three youngest sisters talked mostly about playing with their sibling or everyday interactions. Their statements about their sibling's illnesses were limited and reflected uncertainties. The thirteen year old did lots of work to help me understand her brother Jagger e). The four sisters in their mid twenties gave insights into how siblings react as they grow up with responsibilities for caring and one said that when your 'sister' is ill for a long period of time you realise early on that you only have two choices,

> *"Accept it and adjust or rebel."*
> *Imogen c) - sister.*

4.2 Conceptual cases: unfolding life stories

I have created a series of conceptual cases to illustrate more about the life stories of participating children, without revealing real individuals. All of the conceptual cases are based on combined stories from two or more families and some children and their families contributed to more then one conceptual case. Single and multiple case analysis revealed that the children's life stories (as told by the children and or their family members) followed a range of similar patterns – for example the life limiting illness emerged at birth or later in childhood and were structured around critical moments or changes in the child's state of health caused by the life limiting illness. The conceptual case have been created by using information about the similarities and differences in the patterns of participants life stories and to highlight critical moments for families when the life limiting nature of the illness became evident – a concept I have called 'moments of realisation'. The conceptual cases provide an introduction to how life limiting illness can emerge and impact on children and their families. The theoretical concept 'moments of realisation' – emerges in these stories but is explored in more detail in the following chapter.

Despite the wide range of medical conditions and variety of physical, emotional and social needs of children affected by life limiting conditions, the conceptual cases highlighted how common elements in the life stories of the children and families who took part in this research formed similar patterns. Therefore, the conceptual cases suggested models for understanding the unfolding impact of the child's illness on whole families. Models suggested by the conceptual cases do not describe the entire course of life limiting illness from diagnosis to death but offer a series of complex and multidimensional 'portraits' that illustrate more then the linear passing of time.

I have used the conceptual label – 'moments of realisation' to highlight times of crisis for the children and their families when the threat to the child's life became especially clear to families. These moments ebbed and flowed throughout the child's life in different and often unpredictable patterns. The conceptual cases serve dual purposes. Firstly they provide windows into the lives of life limited children and their families and secondly they begin to suggest the concept of 'moments of realisation', which may be useful in thinking about the needs of life limited children and their families.

'Moments of realisation' formed similar patterns across multiple cases and although, as Table 6 shows, 'moments of realisation' were not always exclusive to the cases used to create each conceptual case, I did try to combine cases where the children's life stories were most similar. Table 6 reveals how 'moments of realisation' appeared across actual cases, which are shown across the top of the table combining names drawn from the conceptual cases with a letter to denote real individuals. A ✓ indicates where specific 'moments of realisation' were used to create the named conceptual case. A ☑ indicates although particular 'moments of realisation' were relevant to a real case they were not used within the named conceptual case.

Table 6 How 'moments of realisation' appeared across cases

'MOMENTS OF REALISATION'	CASES										
	Reuben a)	Reuben b)	Imogen c)	Imogen d)	Jagger e)	Jagger f)	Jagger g)	Daphne h)	Leonora i)	Marcus j)	Marcus k)
Early diagnosis	✓ 'Reuben'	✓ 'Reuben'				☑		☑	☑	☑	
Life threatening surgery	✓ 'Reuben'	✓ 'Reuben'									
Childhood illness			✓ 'Imogen'	✓ 'Imogen'	☑						
Acute loss of abilities			✓ 'Imogen'	✓ 'Imogen'							
Slow deterioration					✓ 'Jagger'	✓ 'Jagger'	✓ 'Jagger'			☑	☑
No diagnosis					✓ 'Jagger'		✓ 'Jagger'				☑
Questions of inheritance						☑	✓ 'Daphne & Leonora'	✓ 'Daphne & Leonora'			✓ 'Daphne & Leonora'
Cycle of crisis & survival		☑	☑	☑			✓ 'Daphne and Leonora'	✓ 'Daphne and Leonora'	✓ 'Daphne and Leonora'	☑	✓ 'Daphne and Leonora'
Dying the next step										✓ 'Marcus'	✓ 'Marcus'

✓ Indicates where specific 'moments of realisation' were used to create the named conceptual case.

☑ Indicates although particular 'moments of realisation' were relevant to a real case they were not used within the named conceptual case.

4.3 Reuben

4.3.1 Early diagnosis and life threatening surgery

The life stories of two children were used to create the conceptual case Reuben. Table 7 provides information about the children's age groups, ability to communicate, number of siblings and the process of research with the families.

Table 7 Reuben: infotmation about contributing children and families

Case	Child's age group	Child's communication	No of siblings	Time with family	Meetings and settings	Ways of working
Reuben a)	2-6 years	Verbal	3	4 months	8 Home Total 8	Talking Drawing Books Family photos Taking family photos Writing Spider chart Story and identity described by family Participant observation
Reuben b)	7-11 years	Verbal	3	2 months	2 Home 3 Hospice 1 School 1Work Total 7	Talking Drawing Books My news poster Presentation re visiting hospice Photos of visit to hospice Story and identity described by family Participant observation

Reuben was born with the severe form of a rare condition that can affect different organs of the body, causing complex heart defects and liver disease in the case of Reuben. Reuben had undergone various operations on his heart that were palliative rather then curative and a liver transplant. Reuben had slight developmental delay caused by his condition. At the time of the study Reuben was six years old and lived with his parents, brother aged 12 (who suffered from Attention Deficit Hyperactivity Disorder ADHD) and sister aged four. Reuben's older sister (aged 19) had recently left home and was living in a flat in the next village. Reuben's father worked shifts as a security guard. The family lived in a four bedroom semi-detached house in a semi-rural area close to a small town. The house was always immaculate whenever I visited.

Reuben's mother described how she went for a routine prenatal scan when she was pregnant with Reuben and had no idea what news she was about to hear or any reason to fear. In the words of the radiographer this mother felt her unborn child move from the status of '*your baby boy*' to '*the foetus*' as she was told that the baby's heart looked '*awful*'. This kind of unanticipated shock was reflected in the stories of other parents, for example in one family, where blood tests were proof of no problem for one child and evidence of life limiting illness for another.

Having been thrown into a nightmare, Reuben's parents quickly found that very little was known about their son's diagnosis. Printed information provided at the time of Reuben's birth covered such a wide range of scenarios about how children with his diagnosis might progress that even when supported by careful explanation it left the parents more bewildered than informed. Reuben's father felt that having a diagnosis provided a label for accessing support services, although little was known about the condition. When Reuben's family telephoned the General Practitioner for support the doctor said she had not heard of the condition but told them, '*By the time you get here I will*'. When they attended for their appointment three days later the doctor produced paper copies of information she had found on the Internet and Reuben's father said, '*We went snap!*' The diagnosis was so rare that the parents and doctor were reliant on the same source of information. It seemed from other cases that the combination of Internet technology and paucity of information about so many life limiting childhood illnesses can combine to create these moments when families and physicians have equal access to knowledge. Perhaps these were moments when authority between doctors and 'patients' was equivalent but sadly everyone was depending on a small pool of information.

Reuben underwent two operations on his heart as a baby and by the age of three surgeons suggested that he needed a liver transplant. Reuben's whole family underwent an assessment to decide whether Reuben should go on the waiting list for a liver transplant. Reuben's father described how he and his wife went on different emotional journeys during the assessment week. However, they came to the same conclusion, that a liver transplant would give Reuben his only chance of a better quality of life. Reuben had little to say about his liver transplant except that he did remember having it. However, his mother doubted this because he was only three at the time. Reuben's mother felt that he muddled up all the various trips to hospital, for blood tests, check ups and major surgery. It was difficult to tell because Reuben only talked about his illness in terms of it being part of his life through events that he described as routine - for example, *"I've been to hospital hundreds of times"*. Reuben and all members of his family talked about the need for repeated major surgery. His parents disagreed about further operations and Reuben and his siblings had not been told that Reuben was suffering chronic rejection following his liver transplant or that the consultants had mixed ideas about further heart surgery.

All members of Reuben's family had their own personal needs. For example, Reuben's father had asthma and his brother suffered from ADHD, but Reuben's needs generally demanded immediate attention. Reuben's mother suggested it would be simpler not to plan ahead but she loved all her children and wanted Reuben and his siblings to enjoy a range of experiences. Each child went to at least two after school activities, one for themselves and the other with their siblings. However, this was in the context of repeated planned and emergency hospital visits for Reuben. Reuben's parents said that daily life for their family was a sort of tight ropewalk in 'limbo'. The life limiting nature of Reuben's illness and constant threat to his life placed the whole family in an intermediate place where even the simple details of everyday life could not be taken for granted.

Reuben's sister (aged 19) felt that compared to other siblings of seriously ill children her parents made sure that she and her brother (aged 12) took on hardly any extra responsibilities in terms of caring for Reuben. However, she felt that her parents had high expectations for her to achieve things that would be out of reach for Reuben.

Reuben's family talked about how Reuben's needs and their family's needs were misunderstood because Reuben looked relatively well. The feeling of being misunderstood socially and professionally because the child looked so 'normal' was unique to the two cases used to construct this conceptual case. These were judgements arising from the hidden nature of the child's illness. This was especially painful to family members for whom the child's frailty was all too clear. Other children and families who participated felt judged because they looked 'different'.

In this conceptual case, Reuben's family experienced moments when other people - even professionals in hospices - failed to understand Reuben's needs and his parents occasionally felt they had to justify their family's needs for respite even to hospice services. The family did value the support provided by hospices but struggled with social ideas about meanings of the word 'hospice' and notions that using a hospice meant the child was dying, or that the family was in some way losing hope by accepting referral to a hospice.

Predominant themes of the life stories of children used to construct the conceptual case Reuben were the diagnosis of a life limiting illness prenatally or within the first few months of life. Families who were anticipating the arrival of a 'normal' healthy baby were faced with the stark prospect of caring for a newborn baby who they were told would only survive the first few years of life. Families talked about the pain of getting 'close' to the child and seeing the child develop but knowing they would not live into adulthood. One mother likened their family life to *'living on death row'*. The threat to the child's life was immediate and on-going but the children were given extra time with repeated major but palliative surgery and this was the second central theme for children and families illustrated through the story of Reuben. Although surgery brought hope for relief of symptoms and better quality of life for the child and extra time to live, it did not represent cure for any of the children taking part in this research. In terms of time, these children outlived all early medical expectations but the length of their lives could still not be determined.

4.4 Imogen

4.4.1 Childhood illness and acute loss of abilities

The conceptual case Imogen is based on the life stories of two children and Table 8: provides details of these children and work with their families.

Table 8 Imogen: information about contributing children and families

Case	Child's age group	Child's communication	No of siblings	Time with family	Meetings and settings	Ways of working
Imogen c)	12-18 years	Verbal	2	3 months	5 Home 1 Hospice 1 Coffee bar Total 7	Talking Dictaphone Drawing Poems Clay modelling Family photos Church newsletter Story and identity described by family Participant observation
Imogen d)	2-6 years	British Sign Language (BSL)	0	2 months	4 Home 1 Day out from hospice Total 5	BSL translator and video of sessions Books Talking Story and identity explained by family Participant observation

When I met Imogen she was 14 years old and lived with her parents and older sister (aged 24). The family home was a new council house, purpose built for a disabled person and their family. Imogen's mother originated from Africa and her father from the United Kingdom. I was at her home talking to her father when Imogen arrived back from school and we were introduced. She was full of gossip and chat about her day and we all giggled lots as soon as she was through the door. Throughout our work together Imogen was always keen to tease me and could turn most things into a joke or a game. I was astonished when I listened to the tape recording of our first session to find that Imogen's laugh was almost inaudible. It shocked me because we laughed together so much and I never noticed how weak her voice was; a sign of her physical frailty that I missed. I listened over and over to see if it was just a fault in the recording but when I sat down to think about it I realised that Imogen was both consciously and effortlessly a strong presence. Imogen was a person who wanted you to know who she was beyond her disabilities and who made every effort not to talk about her differences. Alongside this she had a natural ability to draw you in so that you barely noticed her fragility, initially.

When Imogen was eight she caught what seemed to be a virus affecting many of her school friends. Imogen and her friends had headaches, high temperatures, vomiting and diarrhoea. Imogen's peers at school recovered from the early symptoms in two or three days. However for Imogen and her family this was the beginning of a serious illness, which over the course of the following year left her unable to walk or see. The family made repeated trips to the general practitioner and tried all the recommended treatments. After several weeks like this Imogen's parents took her to the accident and emergency units at their local hospital in desperation there Imogen's parents felt as if care professionals did not take their daughter's condition seriously and that their fears were unheard. Even though Imogen was admitted to the children's ward, her parents were met with what they referred to as *'complacency'* and *'no sense of urgency'*.

Imogen's parents like others reported a pivotal moment when one professional finally challenged the status quo. A nurse on the morning shift arrived to find Imogen covered in sweat and demanded of the night staff *'What is going on?'* In this moment the child's illness seemed to be defined as critical in medical terms and this was apparent to the parents in the sudden response of professionals. Imogen's mother described how the consultant was called and suddenly she said it was, *'anti-this drip anti-that drip!'* Imogen's parents observed that on occasions professionals only 'hear' and respond to each other.

Imogen and her family were told that the condition she was suffering from was normally found in adults and was rare in minority ethnic groups. In many ways she was an exceptional case. Imogen's parents were told that her condition was one that was being studied but definitive information was limited. Imogen's father said their first question was *'is it genetic?'* but the doctors were unable to give him a clear answer and only talked about indications that the disease might have genetic patterns.

When I met Imogen's family they talked about her medical care as if the best was being done for her. However Imogen's parents were critical of the lack of community based support available to them from social services. Imogen's mother said that overall she felt she had to fight for *'every goddamn thing'* and that information about services was of poor quality and not readily available to parents.

Imogen and her family barely made any reference to the prospect that she might lead a short life. Imogen's illness left her unable to walk or see and in her own accounts altered the identity she had previously begun to develop. Imogen was successful in athletics and it frustrated her that her physical education lessons consisted of *'stretching'* and *'messing about'* with a sound ball. Much later in our work together Imogen revealed that her worst moment was when she learned that she would probably not walk again; in that moment she lost her identity as an 'athletic gold medal winner.' Imogen made very little reference to her loss of vision except to tell me that it is called *'visual impairment'* and she could see light as well as using all her senses to take in her surroundings. Again she referred to positive support for people with visual impairment - for example, facilities at the local cinema, which enabled her to listen to descriptions of the film and enjoy time out with her friends.

In terms of day-to-day living Imogen felt that, apart from her parents, her best source of support was her sister who was helping with Imogen's care on a daily basis. Imogen's sister said that for her, helping with Imogen's care felt natural especially to begin with. However, six years on, Imogen's sister admitted that she had to adapt and miss out on some experiences that would have been part of her teens and early twenties. Imogen's sister struggled with the pain of loving her sister and being aware of all the opportunities Imogen had also lost. Imogen's parents struggled with similar painful emotions and her father illustrated their agony when he said that Imogen's spirit had returned after the initial illness, but she was *'a different kind of lovely'.*

Central to the life stories of the children and families contributing to the conceptual case of Imogen was the fact that the life limiting illness did not materialize until after the first five or ten years of the child's life. The children contributing to this story were developing 'normally' until the onset of their illness but the child who was emerging disappeared or changed forever with the onset of the illness. The immediate threat to the child's life came with the acute onset of the illness but was tied to intense uncertainty about whether the child would survive the acute phase of the illness. Families talked about the terror of suddenly facing the possible death of a child who had previously seemed healthy. Following the child's survival from the acute phase of the illness, families were initially greatly relieved but realised over time that the child had residual loss of motor skills or suffered permanent sensory impairments, such as loss of hearing or vision.

With the passage of one year and then many years the children whose stories have been combined in the conceptual case of Imogen seemed, in their families' accounts, to have passed the critical stage of the illness. Where their overall state of health and abilities did not decline any further, families stopped talking about the threat to the child's life. These children and families focused in the longer term on getting support for the child's on-going physical needs in terms of - for example, house adaptations and equipment. Although these children used the hospice service, neither the children nor family members made any reference to the child's frailty or vulnerability beyond obvious disabilities. The child's life expectancy was not central to the stories of these families and it was as if the threat to the child's life was confined to the acute phase of the illness. This was, however, challenged by recurring symptoms and future acute episodes of the illness, at which times the family members became less certain about the stability of the child's state of health and whether they might still die young.

4.5 Daphne and Leonora

4.5.1 Questions of inheritance, crisis and survival

The life stories of four children from three families were used to construct the conceptual case Daphne and Leonora and information about these four children is provided in Table 9.

Table 9: Daphne and Leonora: information about contributing children and families

Case	Child's age group	Child's communication	No of siblings	Time with family	Meetings and settings	Ways of working
Daphne h)	12-18 years	Verbal	3	4 months	5 Home 1 Telephone Interview Total 6	Talking Photos Drawing Books My news poster Story and identity described by family Participant observation
Leonora i)	2-6 years	Non-verbal	0	2 months	1 Home 1 Hospice 1 Work place Total 4	Story and identity explained by family Photos of visit to hospice by care team member and researcher Participant observation
Jagger g)	7-11 years	Non-verbal	2	4 months	4 Home 1 Hospice 1 Family day out 1 Coffee bar Total 7	Story and identity explained by family Participant observation
Marcus k)	12-18 years	Non-verbal	2	4 months	4 Home 1 Hospice 1 Family day out 1 Coffee bar Total 7	Story and identity explained by family Participant observation

Leonora, aged four and Daphne, aged 17 were sisters affected by the same rare inherited life limiting condition. The condition affects children differently and Leonora had a more severe form of the disease then her older sister. Leonora was unable to communicate verbally and her family described her. Daphne who had learning difficulties and developmental delay was able to speak for herself. Daphne and Leonora lived with their parents and brother aged 14. Their brother decided not to take part in the research. Their sister aged 24 had always taken a lead role in caring for Daphne and Leonora. Although their sister had moved to a flat near the family home, she was a daily visitor and continued to help with caring for Daphne and Leonora. Daphne had a room of her own downstairs and the house had no adaptations when I met the family. Their father worked as a business consultant and their mother cared for the children.

When Daphne was born her mother felt that the medical and nursing team struggled to find a way to explain to the family that something was wrong. Looking back, Daphne's mother felt that the doctors were hoping she or her husband would notice signs of her disability themselves and ask questions. However Daphne's mother explained, '*We are simple people and we was very young.*' Daphne's father said that eventually a specialist nurse arrived with an information leaflet and told them that their baby had an *'abnormality'*.

As Daphne grew up it became clear that she did not have a severe form of the medical condition. Her mother found that although Daphne was slow to walk and talk, she needed hardly any more attention than her older sister. Their son was born with no problems and then Daphne's mother unexpectedly fell pregnant again. A prenatal scan revealed that the baby was affected by the same medical condition Daphne had and a doctor asked the parents whether they wanted to terminate the pregnancy. When Leonora was born it was immediately clear that she had a more severe form of the condition than Daphne and her mother said, '*We seemed to live in ICU for months. Almost at the same time Daphne took worse and we were travelling back and forth to the hospital all the time.*'

Their father expressed his love and dilemma through the notion that what families need is not just the child's diagnosis but some idea of potential long-term consequences for the whole family. However he was aware of the difficulties for medical and allied professionals who have to impart this kind of information.

Their parents went on to describe the many moments when Daphne and Leonora had come close to death. Daphne and Leonora had both experienced and survived many critical episodes caused by their medical condition. Their father explained that he felt he had lost Leonora many times and found it more and more painful to get close to her, knowing that she could still die or be close to death again. He had made the decision to focus his attention on work, a lonely and difficult choice to make. Their mother coped day-to-day by focusing her attention on the child with the most needs for care.

Daphne and I met three times and she decided that I should write down what she told me about her life as we worked together. Daphne described herself and her life in her own words. Like all of the young people who could speak to me, Daphne began with the things that were most important to her; she was a seventeen-year-old student with a boyfriend. Her descriptions of visits to the hospital highlighted that she had lots of experience and medical knowledge linked to her experiences despite her developmental delay - for example, she had a simple understanding about the heart and circulation of blood through the body. In terms of her experiences Daphne explained that being in hospital was quite often boring but ambulances were frightening. Daphne dreamt of one day having her own family and becoming a nurse when she would tell sick children what was happening to them. Daphne's mother was aware of Daphne's longing for some independence from the family but was unable to find ways to support her wishes.

Daphne and Leonora's father was especially concerned about the impact of day-to-day care for Daphne and Leonora on their brother and sister. He felt that they had been sidelined at times and regretted that their older sister was so involved in caring for Daphne and Leonora that she seemed to have no life of her own. Daphne and Leonora's older sister was very reserved with me in the family home although she had expressed an interest in participating in the research. Eventually she asked me to telephone her one evening in her own flat, which I did. Whilst their sister was able to express some early memories of resentment towards her siblings, she had become a primary carer and said this was because of her own genuine interest in people with special needs and not solely related to caring for her sisters.

Where the child's condition was inherited or familial this overwhelmed the accounts of parents contributing to the conceptual cases of Leonora and Daphne. In these cases the moment of diagnosis was described but the primary theme for parents was not just the fact of the medical diagnosis but the personal struggle of facing issues related to the genetically inherited nature of the child's illness. No child or sibling made any reference to the fact that the child's life limiting illness was inherited nor did older siblings talk about the implications for them of having their own children in the future. However, parents expressed feelings of personal guilt, responsibility and blame, even though they had no way of knowing that their child would inherit a life limiting illness. Parents' fears also extended to siblings and future generations of children who might also inherit the same condition. The issue of inheritance was not linked to time in the accounts of parents. Although parents talked about the moment they were given the child's diagnosis as a specific event in time, issues related to the inherited nature of the child's condition pervaded many themes and was not confined to one account of the moment of hearing that the illness was genetically linked.

To some degree all of the children taking part in the research had faced more then one critical episode in their illness. However for some children these phases of the illness brought a decline in health, deterioration of skills or loss of abilities but not the immediate and repeated threat to the child's life faced by the children and families contributing to this conceptual case. These families were forced to confront the fact that the child could die at any moment on many occasions but paradoxically repeated past experience told them that the child could be remarkably resilient because they had survived so many moments of 'nearly dying'. Therefore certainty and uncertainty about the child's survival fluctuated amongst families facing this aspect of the child's life limiting illness and fears were pervasive.

4.6 Jagger

4.6.1 Slow deterioration and no diagnosis

The life stories of three children contribute to the conceptual case 'Jagger' and Table 10 provides details of these children and work with their families.

Table 10: Jagger: information about contributing children and families

Case	Child's age group	Child's communication	No of siblings	Time with family	Meetings and settings	Ways of working
Jagger e)	7-11 years	Non-verbal	1	5 months	5 Home Total 5	Story and identity explained by family Mind map Family photos Family video Talking Taking photos Magnetic poems Poster Video by sister Participant observation
Jagger f)	2-6 years	Non-verbal	0	3 months	2 Home Total 2	Story and identity explained by family Talking Drawing Participant observation
Jagger g)	7-11 years	Non-verbal	2	4 months	4 Home 1 Hospice 1 Family day out 1 Coffee bar Total 7	Story and identity explained by family Participant observation

When I met Jagger he was nine years old. He lived with his mother, sister aged 12 and little brother aged two. The house was a small council property in a street of identical houses. The paint was peeling off the door and inside was always chaotic and muddled. The house had been adapted for Jagger with the addition of a downstairs bedroom and overhead hoists. Jagger's mother told me once that in an ideal world she would be a minimalist, laughing when I smiled at her, she said, *'Well I know ... I do know!'* Jagger's mother said she felt it was important to take part in my research, to share their experiences and maybe help other families. She also wanted to give Jagger's sister the chance to talk about her own story in private with me and get a chance to really say what it was like for her that her brother had disabilities. These aims were common to other families who participated in the research. Jagger's mother used family photographs and video to illustrate his story to me. Her daughter sat next to me to listen and look. Jagger sat with us in his usual seat on the sofa opposite the television. Occasionally Jagger would vocalise and move his arms and legs. Jagger responded most to his family. His mother and sister went to him many times talking to him and kissing him.

When Jagger was born his mother's only concern about him being premature was that he was tiny ... *'a bag of sugar.'* She talked of developmental delay but distinguished this from disability. Jagger developed normally until he was three years old and then a series of long lasting epileptic seizures left him with profound disabilities. Jagger's underlying medical condition was undiagnosed. Jagger's mother began with a series of family photographs showing Jagger's first three years of life. He looked animated, playful and as if he were growing and developing new skills. Aware that the three of us were absorbed in the moment of celebrating Jagger's early life, development and achievements, Jagger's mother paused and prepared us both for what came next. Closing the photograph album for a moment, Jagger's mother paused and told me to get ready for a shock. She turned the page and there was a photograph of Jagger sitting in a chair with support straps around his waist. His head was slumped forward and it could easily have been a different child. Jagger's mother explained that at the age of three Jagger started to have major epileptic seizures. All the medical tests were negative and over four or five months Jagger lost all of the skills he had gained.

Jagger's mother struggled to find words to describe the changes in his abilities referring instead to more photographs in which Jagger looked like the same almost lifeless child in a series of different locations. She had lost the child who was, *'developing lovely'* and in his place was her vulnerable nine-year-old son with hardly any more skills then a newborn child. Jagger's mother couldn't find photographs to illustrate their daily life. Day-to-day Jagger's mother felt that his care needs were her responsibility, even though she and her daughter struggled to cope with the number and range of professional carers who, *'breezed in and breezed out.'* Not only did she refer to responsibility for all of Jagger's basic physical needs, she expressed the burden and guilt of constantly having to assess whether changes in Jagger's epilepsy or respiratory problems required further effort from herself or medical attention. Jagger's family did not want more 'carers' involved in their lives they needed individuals who could somehow meet their needs as a family.

Jagger's mother explained that she rarely asked for help and in the first year or two after his condition deteriorated she was afraid that one or all of her children might be taken into care. Jagger's mother was a lone parent caring for two other children beside Jagger.

Parents in families with more then one child expressed this practical and emotional dilemma of how to priorities and respond to the needs of all their children in a variety of ways. This might be common to all families with two or more children but for the families who took part in this study the needs of one child (the child affected by a life limiting condition) frequently outweighed the needs of siblings. Finding a recent photograph of Jagger and his sister looking into each other's eyes and smiling, Jagger's mother talked about losing her son and how she now looks for little moments to treasure.

The story of Jagger illustrates that not all families caring for a child they have been told has a limited life expectancy ever know the child's medical diagnosis. The children and families contributing to this conceptual case focused on the child's early development, which did not follow developmental norms but was constructed by parents and older siblings as satisfactory given the child's disabilities or *'fine for him'*. At some point in the child's life story, however, deterioration emerged through loss of developmental achievements: *'He lost everything he could have done before.'* This was through neurological damage in the cases of study participants. Although parents were told that the child would die young, they had no certainty about how changes might appear or when they might arise. These parents hoped that a name for their child's medical condition would provide them with some evidence or at least the chance to seek out facts that would illuminate something about the likely path ahead for the child and family. Families contributing to the conceptual case of Jagger expressed some doubts about the life limiting nature of the child's illness given that the medical diagnosis remained unnamed.

4.7 Marcus

4.7.1 When dying is the natural next step

The conceptual case 'Marcus' represents the stories of two children whose parents had been told their child would be unlikely to live for more then one or two years at the most. Table 11 provides information about the children and their families.

Table 11: Marcus: information about contributing children and families

Case	Child's age group	Child's communication	No of siblings	Time with family	Meetings and settings	Ways of working
Marcus k)	12-18 years	Non-verbal	2	4 months	4 Home 1 Hospice 1 Family day out 1 Coffee bar Total 7	Story and identity explained by family Participant observation
Marcus j)	12-18 years	Non-verbal	1	6 months	4 Home 1 School Total 5	Story and identity explained by family School work teacher School communication aids photo researcher Participant observation

When I met Marcus he was unable to move purposefully or independently, he was unable to communicate verbally, he often looked exhausted and barely made eye contact. I also met Marcus on a family day out and at school and found that on more energetic days Marcus could communicate his basic likes and dislikes. Marcus' father made the point that most professionals interact with children like Marcus in one setting for short periods of time, which can make it difficult to see the whole person, as their family see them. When I met Marcus he was 17 years old. He lived alone with his parents and was their only child. Marcus' family chose not to use images or family photographs to illustrate his story or their experiences with him, instead they encouraged me to meet Marcus in a couple of different settings.

Marcus was born with profound disabilities making him prone to severe epileptic seizures and respiratory infections. Marcus' mother talked about being a mother for the first time, the uncertainty of suspecting that something was wrong with her child and the pain of facing the truth. In her account she fluctuated between the young first time mother who she was, longing to feel that her child was developing and *'doing everything he should have been doing'* and the woman she has become, who looks back and knows that Marcus had problems that were difficult to face from the moment he was born.

In the two years before I met Marcus his mother explained that his deterioration in health had been mostly gradual and difficult to see at close hand. Then a doctor told Marcus' parents what they were intuitively aware of but had not heard put into words, that their son would probably only live another year or two. Marcus' mother said that at first it was like someone *'speaking the unspeakable'* but having watched Marcus' condition deteriorate over the months his mother began to feel she could see a moment when dying would be almost the natural next step.

Marcus' parents barely commented on his condition when I was in their home and Marcus was clearly having a bad day. He might be asleep on the sofa or sitting motionless in his wheelchair with his head down. They talked about who Marcus was on a good day, his changing needs and how they wanted to ensure that he enjoyed every moment through a range of experiences. This focus had become the meaning of his mother's own life. However Marcus' mother also knew that a time could come when she might no longer have the physical or emotional resources to face all the ever-increasing challenges of caring for Marcus. Marcus' mother talked of getting the support they needed as a family *'the devious way'* she said they did not shout, they went to people in positions of power and sought their assistance, ability or influence. However Marcus' mother also expressed the idea that she would sometimes like to scream and force professionals to think about life from her son's perspective or stand in her shoes.

Marcus' parents were at different emotional places in terms of facing the news that he had only got a short time to live. Their ability to talk about his death was in stark contrast to each other. Marcus' mother talked a lot about her son's death but there was still evidence in her account that she was no more ready then her husband to accept that Marcus' would die, until she felt that letting him go would be the last and best thing she could do for him. She talked of Marcus teaching her what life was about. Marcus' father communicated very differently to his wife. He did not want to talk about what he called, *'this death thing.'* Marcus' father summarised the views of other parents (not just other fathers) when he said, *'I think about my child dying every waking moment but I can't see how talking about it helps.'* Marcus' mother like other partners waited for the moment when her husband was ready to talk. Marcus' father told me how his experience and memory of his mother's death were linked to her care at an adult hospice. Whilst that support was good and he could see that the facilities at the children's hospice were good … *'deep down inside'* he was not ready to face Marcus' death.

The dominant theme of parents and siblings who had been told that the child probably only had a year or two to live was the child's short length of life and opportunities for them all to experience life to the full. In both cases the children's mothers talked and explored the notion that the child would die, when the moment of the child's dying would come, what symptoms the child might suffer and how they and other family members might react. The fathers on the other hand spoke about the fact that the child would die but did not explore their personal fears, stating the idea that talking was not helpful and would not change what they knew was going to happen. These families were certain that their child would die relatively soon but did not define the child as a dying person at the time of the study. Therefore I cannot be certain when the moment of certainty about a child's dying arrives for families.

Chapter Five

5 Theoretical concept: 'Moments of realisation'

5.1 A developing child at the centre

In every case, children, parents and siblings placed the child's skills, abilities and personality at the heart of their work with me (in talk, drawings, photographs and in settings chosen for participant observation). This was especially true in families where the children were less able to represent themselves. Where I could not hear directly from the child, parents and siblings focused their efforts on helping me to know the child as a unique person with likes, dislikes, personality traits and skills, no matter how small or invisible to outsiders. Development of the child's abilities and individuality was integral to every interaction between the child, siblings, parents and myself. The course of the life limiting illness shaped the developing child: it impacted on their life, understanding of themselves and how they presented themselves to others. To have any understanding of life limiting illness and the impact on the child and their family, one must always remember that children are developing human beings.

5.2 Questions of inheritance, diagnosis and prognosis

Almost all of the participating parents began their accounts of the child's illness with the moment of diagnosis or the moment they were told that even though a diagnosis could not be made the child was unlikely to live into adulthood because of relentless deterioration or because they had become particularly frail and would be susceptible to complications like pneumonia.

The conceptual cases illustrate how diagnosis was prenatal or soon after birth in some cases *[Reuben a), Reuben b), Jagger f), Daphne h), Leonora i) and Marcus j)]*; came later in the child's life for example after a prolonged illness followed by investigation of symptoms over time *[Imogen c) and Imogen d)]* and where no diagnosis was ever reached *[Jagger e), Jagger g) and Marcus k)]*. The child and family's interpretation of the illness was deeply affected by the moment of diagnosis in relation to time in the child's life.

Where diagnosis came before or soon after birth, parents talked as if the future they had anticipated for their child suddenly vanished from view, at the moment of hearing the child would not live into adulthood. Often it seemed that the dissipation of parents' hopes for their child's life were tied to the words of professionals. In these moments unborn or newly born babies no longer represented parents' aspirations and hopes for the future but they were still children with immediate needs. In their accounts of early diagnosis, parents quickly shifted from the shock of diagnosis to focus on the child as a person, whose life might be short, but a child who was valued by their family.

No child mentioned having an unnamed illness. Parents who had been told their child was life limited, although the underlying condition was undiagnosed, sought a name for the threat imposed on their child's life. Parents caring for children with no diagnosis expressed uncertainty about the child's prognosis, expressing the idea that the two things were generally tied together and questioning the certainty of medical staff that the child would lead a short life. Where the child had complex disabilities and their cognition was difficult to assess, parents with no name for the child's medical condition described the child's life story including the concept that the child might lead a short life and searched for facts and evidence that would either confirm of dispute this notion. Where the child was cognitively able and their disabilities were confined to one or two aspects of motor skills and/or some level or sensory impairment, families rarely talked about the anticipated threat to the child's life, if they had no name for the underlying medical condition and especially where the child's general state of health seemed to become stable over time.

Beyond the profound shock of hearing that their child's life expectancy was limited parents talked about diagnosis in different ways:

1. Medical diagnosis as little more then a label with no information attached, used by support services for assessing who was eligible for support; in this way diagnosis facilitated access to support.
2. Where the child had no diagnosis parents hoped not only for a medical name for the child's condition but that it would clarify the path ahead
3. The idea that families need a long term picture about caring for a life limited child (something more then diagnosis).

Hearing a medical diagnosis or a doctor say that their child's illness would limit their life expectancy to childhood began all family stories about the unfolding of the impact of the life limiting illness, even where parents suspected that the child was seriously ill but felt unable to make their fears heard. Whilst parents and siblings talked about the moment they heard the child's diagnosis from the doctor (in the case of parents) or from parents (in the case of siblings), the children rarely mentioned any moment of diagnosis. Whilst the children talked about the consequences of their illness for themselves and their families and named their condition or used family words to refer to their illness, none of them related the moment they first heard about their illness.

Alongside the 'diagnosis' (whether there was one or not) was the issue of the child's 'prognosis'; uncertain in terms of time for all the participating families (no-one knew when the children might die: they might live for months or even years). In two cases information sent to the hospice by medical consultants after the study was complete, suggested that improvements in the child's health status meant they were expected to live into adulthood and therefore no longer met the hospice criteria for support. In a third case the young person reached her eighteenth birthday and support services including the hospice had to work towards finding transition support for her through residential care and adult services. No parents used the word 'prognosis' but they talked about the uncertainty of how long the child might live, the child's repeated survival from critical episodes or even ambiguity at the moment when the child had seemed very close to death.

Most participating parents made reference to questions about whether the child's condition was inherited or not. Parents expressed fears and painful emotions principally tied to feelings of guilt. However, although parents described discussions about genetically inherited disorders with medical staff, it did not seem in any case to be a separate moment in the course of the child's illness but integral to questions and fears linked to the moment of diagnosis. This was so whether answers about questions of inheritance came soon or were ambiguous. Figure 3 illustrates the interplay between questions of inheritance, diagnosis, prognosis, time and the child's uncertain future.

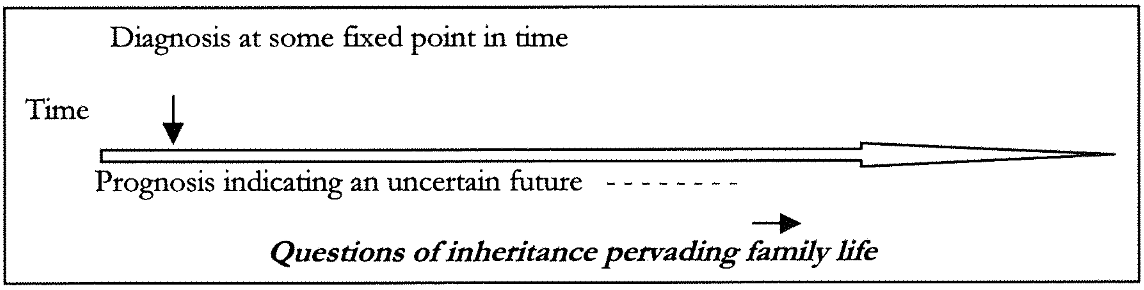


Figure 3 Questions of inheritance, diagnosis and prognosis

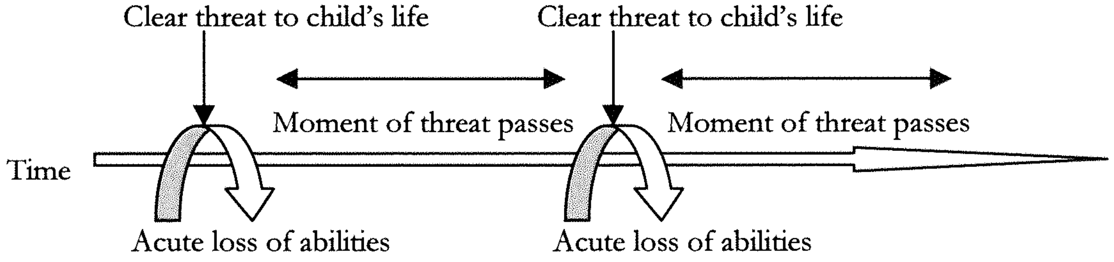
5.3 Acute loss of abilities

Invariably the sudden loss of skills or impairment - for example, loss of hearing, vision or mobility - emerged after critical illness. Children and families talking about acute loss of skills always aligned these points in the child's life story with time. In some cases acute loss of skills was linked to the start of the child's illness but could also be a consequence of critical episodes in which the child's state of health suddenly declined due to - for example, haemorrhage, or extension of brain and or neurological damage.

Acute changes brought certainty about the threat to the child's life to the children and their families. The child's physical frailty, previously less visible (especially for children who were described by their families as charismatic in some way) intruded on the child's and family's sense of the child as a person and brought realisation of the life limiting nature of the child's illness. However, where acute loss of abilities and skills was followed by months or years where the child's condition remained unchanged, time faded perceptions of the immediate threat to the child's life amongst the child and their family.

Acute loss of abilities and skills were therefore always linked to particular events in time. Children and families talked about the moment of realisation that the child's abilities had changed suddenly, but they also reflected back on changes that had occurred months or years before, as if the absence of any further change was a positive sign. Passing of time in this way made the threat to the child's life ambiguous to families, regardless of how complex the child's needs or disabilities.

Whilst sudden loss of skills or abilities was central to the child's life story as explained by family members, they were themed as temporary disruptions, encapsulated by children and families as events that had happened in the past but supported by evidence that the moment of immediate threat to the child's life had passed. Figure 4 illustrates how the threat to the child's life was clarified at times when the life limiting illness caused acute loss of abilities but seemed to fade in between these episodes.



Questions about whether the child will live or die at times of acute change fading overtime until the next acute phase

Figure 4 Acute loss of abilities

5.4 Slow deterioration

Where families described slow deterioration in the child's state of health *[Jagger e), Jagger f), Jagger g), Marcus j) and Marcus k)]* parents talked of not seeing the changes themselves but eventually having changes pointed out by medical staff, allied professionals or members of their extended family.

Slow deterioration in a child's state of health was revealed in physical changes - for example, gradual loss of weight, subtle changes in motor skills and increasing length or severity of seizures: changes that were very difficult for parents to see at close hand when they had to focus day-to-day on meeting the child's care needs. Individuals who were not responsible for the child's daily care therefore noticed changes that emerged slowly, partly because they saw the child infrequently. Parents described enormous pain and guilt at not having noticed changes in the child's health themselves. Despite all the care and love they gave the child, parents blamed themselves when they failed to see changes in their child's body that were in effect invisible moment to moment, but revealed slow deterioration the moment they were noticed.

More than any other element of the child's unfolding story, slow deterioration was a theme described by parents rather than children or their siblings. Parents faced with the realisation that their child's condition had deteriorated, looked back over long periods of trying to meet all the child's needs and wondered why they had not been able to see what they were witness too. This realisation of changes in the child's body over prolonged periods of time clarified the threat to the child's life as immediately as acute changes when others described them to parents. In some ways parental accounts of slow changes in the child's health seemed to generate greater feelings of helplessness in parents, because the disease was revealed as relentless. In these moments parents knew that whatever they did, the course of the child's illness would lead to an early death. Slow deterioration was revealed through physical changes in the child's body. Parents confronted with the fact that their child's health had declined over time talked about **when** the child would die in contrast to parents faced with the immediate threat of acute changes who talked about **whether** the child would die. Slow deterioration in the child's condition seemed to make it clear to parents that the child's life expectancy was indeed limited. However, parents differentiated the notion of a shortened life expectancy from the concept of dying. Figure 5 illustrates how slow deterioration in the child's condition revealed the relentless course of the illness and made the threat to the child's life very clear.

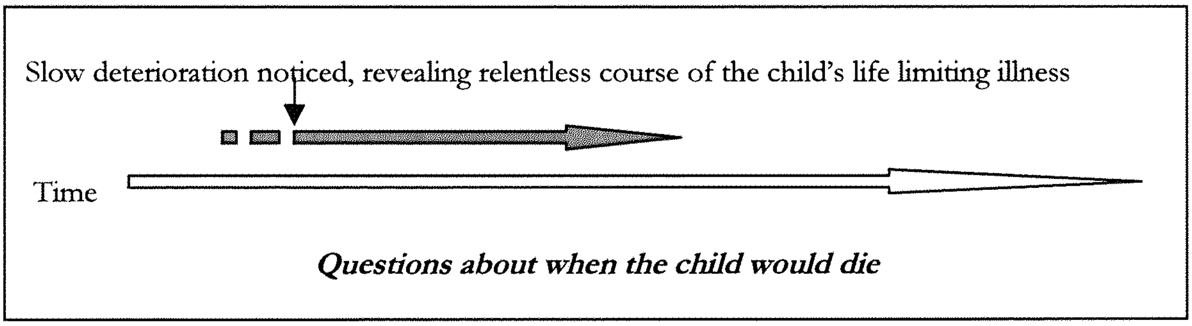


Figure 5: Slow deterioration

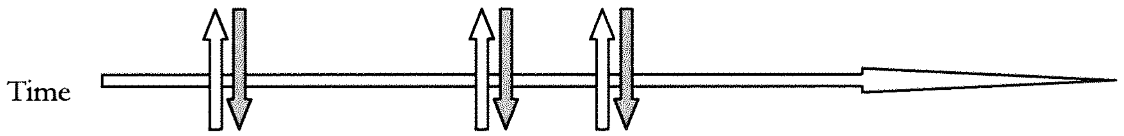
5.5 Life threatening surgery

A minority of children who participated in this study faced repeated major life threatening surgery - for example, open heart surgery or organ transplant *[Reuben a), and Reuben b)]*. Therefore as an element of the children's life stories 'life threatening surgery' may only apply to a small proportion of children. However, where it was relevant to the course of a child's illness, life threatening surgery was a major theme for all family members.

The need for life threatening surgery could come at any point in the child's life and arose more then once for some children. All family members talked about the child's surgery in their own ways: memories of emotions and experiences were mixed but together provided very detailed pictures of the time leading up to, during and after major surgery. Within accounts were elements of how ill the child was before surgery, hopes that surgery would somehow 'fix' the child and result in cure or at least give the child hope for a prolonged life. Families faced disappointment when surgery only provided a little improvement, giving the child a bit longer to live or some improvement in quality of life, although the underlying condition continued to be life limiting. However, the families I met continued to hope against hope that the next operation would provide yet greater improvements.

Life threatening surgery represented major events in the child's life story but was linked to hopes and aspirations for some improvement in the child's quality or length of life. Surgery was revealed through the child and family's accounts and through physical scars on the child's body. Children with uncertain memories of surgery when they were very young used their scars as physical evidence of that which they could not articulate. Parents and siblings used scars to confirm that the child was different and fragile in some ways. Parents also provided young children with explanations for hidden away scars that might be seen socially - for example, in the summer or during swimming. These efforts were confined to simple links to the fact of having had an operation but not to the underlying diagnosis or the life limiting nature of the child's illness. Figure 6 illustrates how repeated life threatening surgery brought hope alongside the threat to the child's life

Repeated life threatening surgery: - Hope alongside the threat to the child's life



Questions about risks and benefits of repeated major surgery from which the child could die

Figure 6 Life threatening surgery

5.6 The cycle of crisis and survival

Most children who participated in this study had survived repeated critical episodes in the course of their life limiting illness *[Reuben b), Imogen c), Imogen d), Jagger g), Daphne h), Leonora i), Marcus j) and Marcus k)]*.

Children, parents and siblings talked about the fear associated with collapse, haemorrhage, respiratory distress and memories of ambulances. For children who experienced a cycle of crisis and survival the threat to their lives became uncertain to family members, illustrated by ideas about the child always being able to 'pull through' or being 'strong' rather than vulnerable.

Family members who talked about the child's unexpected survival from many crises, acknowledged that the child could have died many times. However, they struggled with their own grief and the emotional pain of facing the ever-present terror that the child could die at the next moment of crisis, which could come at any time. Parents who talked about the course of their child's illness in terms of a cycle of crisis and survival indicated that this element of the child's life story especially, set them apart from other children and families in society. Parents were aware that in this society children are expected to reach adulthood and they were even more certain that most parents could not begin to comprehend the experience of caring for a child who 'almost died' many times.

Cycles of crisis and survival made the child's unfolding life story increasingly different from the life stories of 'other' children. Parents and siblings described these episodes for children who communicated non-verbally. Children able to speak for themselves had vague memories of being intubated and of being rushed to hospital following collapse or haemorrhage and linked these memories to times of fear. Figure 7 illustrates that the cycle of crisis and survival faced by some children led their families to construct the idea that the child was 'a survivor'.

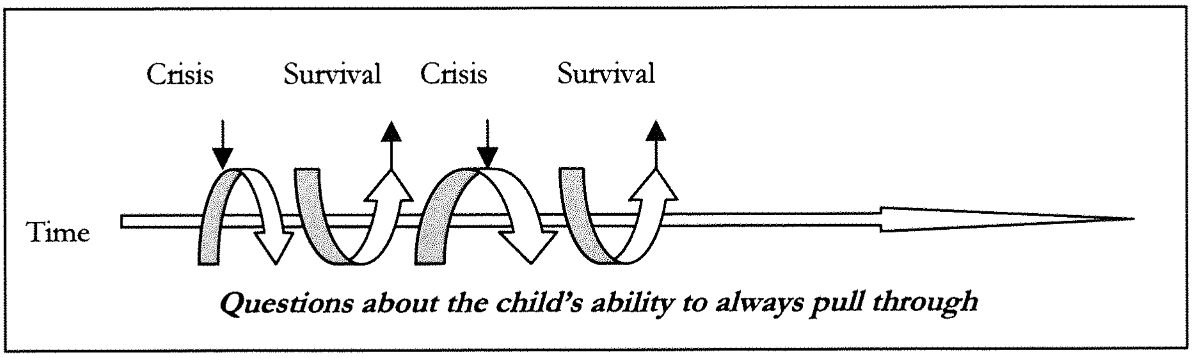


Figure 7 The cycle of crisis and survival

5.7 The child's life and death

The life expectancy of all the children who participated in the study was unpredictable. No-one could be sure how many months or years the children would live. Parents who participated in this study were overwhelmingly searching for the meaning of life and how to help their child make the most of their life – even though it would be short. Marcus' story represents the two children who seemed more likely to die within a year or two than the other children taking part in this study *[Marcus j) and Marcus k)]*. In caring for these children their parents and siblings did not struggle with the meaning of death. They focused on the meaning of the child's life and made every effort to ensure that every moment was special for their child.

I did not meet children who were 'dying'. In this way I have no understanding from the research about the end of life for children who are affected by life limiting conditions. All I did get a sense of was that children affected by life limiting conditions and their families focused on life and living. I do not know when the moment of the child's dying becomes clear to families. However, there was a time when families taking part in this study realised that their child would die within the next year or two but still in the context of the child having time to live. Figure 8 illustrates how families focused on life and living even for children who they knew would die within a year or two.

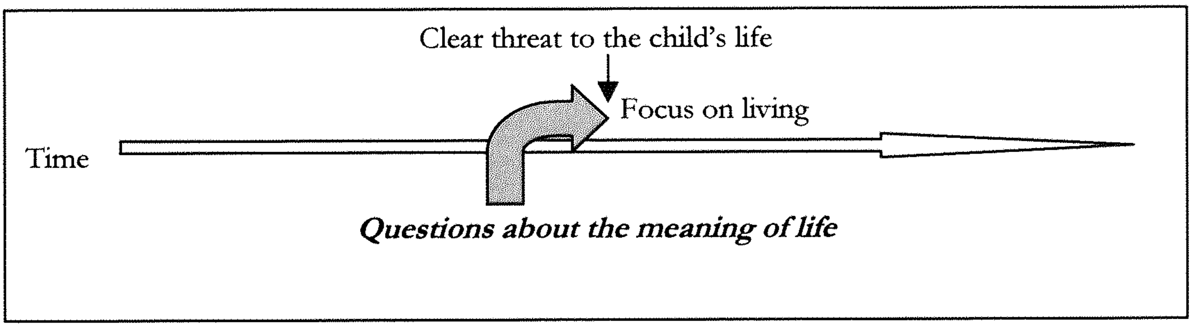


Figure 8 The child's life and death

5.8 Summary

To date there has been no research to reveal the long term, day-to-day experience of life limiting illness in childhood. This study has uncovered common elements ('moments of realisation') in the children's life stories regardless of their diagnosis. These common elements were found to follow similar patterns across cases using Grounded Theory for analysis. The conceptual cases presented in chapter four represent a weaving together of two or more real stories around common elements. To illustrate – merging together two stories with the emphasis on the elements; an early diagnosis and major surgery created the conceptual case Reuben. All aspects of the case come from one or other of the real stories. The conceptual cases suggest new models for understanding how the life stories of life limited children can unfold in similar ways (have 'moments of realisation' that emerge through common patterns). This was despite the wide range of diagnoses.

Overall the conceptual cases illustrate four fundamental points:

i) Being affected by a life limiting condition in childhood is not one experience: the child's life story can unfold in different ways but there are common 'moments of realisation' in the life stories of life limited children

ii) 'Moments of realisation' occurred time after time in the child's unfolding life story and generated feelings of fear and uncertainty for their child and family because the threat to the child's life was made clear at these times

iii) The threat to the child's life affected the day-to-day interactions and experiences of the whole family and each family member

iv) In most cases the child's life expectancy could not be accurately anticipated and so the children and families that I met focused on life even when it became clear that the child had only a year or two to live.

The conceptual cases presented here offer the reader a series of portraits that illustrate the complex and multidimensional lives of life limited children and their families. The models suggested by each conceptual case are complex and describe more then the linear impact of the illness on the child and family. Individual 'moments of realisation' in the children's unfolding life stories have been explored in detail. The theoretical concept outlined suggests that over the child's life time the life limiting illness will create reoccurring moments when the threat to the child's life will emerge and become clear to the child and their family, whether the family talk about it or not.

Presentation of the conceptual cases and exploration of 'moments of realisation' begin to challenge the relevance of linear concepts of dying and illness trajectory over long periods of time (years) and across the life span of children who continue to grow and develop, although they are affected by a life limiting illness; children whose needs are barely described in the care and dying literature. The models described begin to raise questions about whether the notion of 'dying trajectory' might only be relevant in the last days of the child's life. This study has not explored this aspect of life limiting illness in childhood because all of the participants were living (not likely to die within days, weeks or even months).

Chapter Six

6 Family awareness and communication context

The purpose of this chapter is to explore how children and families managed talk and awareness about the child's life limiting illness and to link family awareness and communication themes to the concept – 'moments of realisation'.

All the participants recast difficult encounters, painful emotions, troubled relationships and the absence of practical support in terms of communication about the life limiting nature of the child's illness. All the families lived with the pain of knowing that the child would die in childhood and family members struggled with questions about how to communicate about the child's life limiting illness, whilst trying to maintain daily family life. Awareness and talk about the life limiting nature of the child's illness were not in balance and parents faced profound dilemmas about how to be open to the questions of the children and their siblings. The dilemma for the affected child was created by the tension between knowing the family approach to communication about their illness and the need to understand their own life story. Participants tended to divide the subject of the child's life limiting illness into three parts, probably largely unconsciously. The chapter is subdivided into the following themes because they provide a structure for understanding more about how families tried to manage awareness and talk about the child's life limiting illness:

a) The child's life limiting illness
b) Death in childhood
c) The child's dying.

6.1 Family talk: the life limiting illness

Knowing their child was suffering from a life limiting condition generated awareness and the need for talk but parents did not necessarily have facts, insights or ways to begin talking about something so deeply painful to themselves and those they wanted to communicate with but also protect. This can be related to 'moments of realisation' – *[Questions of inheritance, diagnosis and prognosis]* – times and questions that generated uncertainty, fear, guilt and anxiety about how to talk in families of life limited children.

Many parents felt it was their responsibility to determine what information should be given to the child, siblings and extended family members - not only for the quality and quantity of information but for the emotional reactions of each individual. In their efforts to protect all concerned, parents constantly made complex 'unconscious decisions' about who needed to know about the child's illness and how much detail they should be offered or might cope with.

> *"Over the years we have found not only are we responsible for talking to Reuben, we are responsible for what we tell the other children and how worried they would be, and we are responsible for what was told to other family members and how worried they would be. The point is we didn't always know the answers to their questions, so we had no way to relieve their stress. We decided early on to control the flow of information out because we were just getting bombarded with stuff coming back in again and we thought well you know it was just an impossible thing to do. You can't just keep answering questions you don't know the answer to."*
>
> *Reuben a) – father*

Parents faced the long-term problem of how to communicate with: each other, the child, siblings, extended family and friends. In participating families parents tried to take a clear approach and described decisions that evolved over time, especially about how to talk with the child about the life limiting nature of the illness. Initially, parents described two approaches:

1. The family said they talked about the child's life limiting illness with the child
   [Reuben a), Imogen c), Imogen d), Jagger e), Jagger f), Jagger g), Leonora i), Marcus k) and Marcus j)

2. The family said they tried to control how the child's life limiting illness was talked about with the child
   [Reuben b), and Daphne h)].

Parents of children unable to communicate verbally used 'talk' as a process for comforting the chid. The mother of *Jagger f)* talked to him about his illness as much as the family of

Imogen c) talked with her. However, the communication strategies used by these two families had different objectives. *Jagger f)* was aged five, had profound disabilities but had recently gained some physical skills that his mother was told he would loose again over the next five years. *Jagger f)* was unable to vocalise or demonstrate comprehension. *Jagger f)* was with us when I interviewed his mother. We all sat on the floor to talk and she often addressed her comments to *Jagger f)* and they would make eye contact with each other.

> *"I mean he wasn't the usual age for holding a bottle, you know all them sort of things and he wasn't sitting up then either, couldn't sit up either could you Jagger? That's when they diagnosed what was wrong and told us it was life limiting. But now I mean he is a completely different little fella, walking even … what you can do now hey can't you mate? Amazing you are! They have said he will end up in a wheel chair but you can't think of that all the time. We just get on with each day … don't we mate? I don't fully understand what he knows but just try to talk to him like any other little boy … tell him what's wrong, what's going on … all that!"*
>
> *Jagger f) with his mother*

Parents of children whose comprehension was difficult to assess or who communicated non-verbally *[Imogen d), Jagger e), Jagger f), Jagger g), Leonora i), Marcus k) and Marcus j)]* talked and offered explanations along with physical soothing and care even though the children's ability to understand words was uncertain. The mothers of *Jagger g)* and *Leonora i)* explained that regardless of the child's ability to communicate, as parents they were aware of their children's reactions and any signs of discomfort or fear and wanted to offer comfort and reassurance.

Parents of children who were able to communicate verbally *[Reuben a), Reuben b), Imogen c) and Daphne h)]* faced a dilemma. Whilst they wanted to try to keep their child informed about the immediate (short term) effects of the illness, they also wanted to protect the child from the long-term outlook and the life limiting nature of the illness. These parents had to respond to the active role of the child in communication about the illness; they were talking with children able to perceive, assess their own experiences and ask questions from their own perspectives.

In contrast to *Jagger f)*, *Imogen c)* was an articulate 14 year old and talk within her family about her illness was largely lead by Imogen herself.

> *Toni's field notes from visit to family home of Imogen c) to talk to mother*
>
> *During our discussion Imogen's mother made the following points to underline the importance of talking with Imogen about her illness:*
>
> *i) Imogen was articulate, intelligent and had her own questions about the illness and the impact it had on her life*
>
> *ii) No-one was more aware of the physical consequences of the illness than Imogen*
>
> *iii) Imogen had experiences related to her illness, such as visits to the hospital or hospice, where she might be alone or without her family*

Parents had fears and questions about the best ways to talk to children who could comprehend and were developing. Participating parents wondered how other mothers and fathers talked to their children about the child's life limiting illness and parents searched newspaper and television stories for clues and ideas. Many parents told me they were hoping to get ideas from my own work with the children. In the quote below the mother of *Reuben a)* used an account of a television programme about a child undergoing a liver transplant to explore some of her questions about how to talk to *Reuben a)* about his illness.

> *'What I found useful on that programme actually the day before was that they showed the little girl, I had forgotten this, they showed the little girl that had the liver transplant a few years earlier. They said to her um 'oh you didn't have a well liver did you?' and she said 'no my liver wasn't very well', she is about five or six. Her mum said 'oh what colour was your liver?' She said 'my liver was green' and she said 'what colour is your liver now?' and she said 'it's pink' And I thought they have obviously gone into it a lot more than I had with Reuben because I just said you had a new liver because your old liver didn't work properly but they obviously had gone into the colours of the liver and stuff but I thought oh right, ok I mean it probably doesn't mean an awful lot to her but I found it quite interesting that people go into it different ways"*
>
> *Reuben a) - mother*

Parents wanted to know if there were ways to let the subject of the child's illness come up in everyday conversation. Individual family members discussed their personal fears, questions and lack of resources in terms of finding ways to be open about the child's illness without causing distress to the child. These questions arose regardless of the child's age or the overarching pattern of the child's unfolding life story.

There were two families who expressly stated that they tried to control how the child's life limiting illness was talked about with and/or in front of the child *[Reuben b), and Daphne h)]. Reuben b)* was six and *Daphne h)* was seventeen but she had learning difficulties and her level of comprehension was probably about that of a six or seven year old. The family of *Reuben b)* tried to take overarching control of how his illness was discussed with him within the family and in settings outside the family home such as school.

> *Reuben b) – Toni's field notes from visit to child's school*
>
> *Reuben's teacher chatted informally with me about the research. He talked about how the other children ask, 'What is wrong with Reuben?' stating that Reuben's mother had asked for explanations to be kept very simple. The teacher gave an example of how the other children's questions were answered … 'We say … well you know his heart doesn't work properly and that makes him tired and a bit grey at times". Reuben's teacher said that two children in the class had gastrostomy tubes and how school staff would explain to the other children, 'We might say … you know they have lots of bad tasting medicine and so we just give it to them straight into their tummy because there is lots of it and it tastes bad."*
>
> *The teacher felt the children accepted these explanations. I asked if he felt the accounts they were using would continue to be 'enough' for Reuben and the other children and he said, 'Well they are all getting older now and beginning to ask for more information, it was enough while they were little but I can see a time when they will ask more questions." He explained that whilst Reuben was young he didn't have lots of questions about his illness. He thought for a while and then commented, "I think he may begin to have questions soon though."*

The family of *Daphne h)* said they wanted to control any talk with *Daphne h)* about her illness being life limiting. During my time in the family home I noticed that hospital visits were discussed but the concept of *Daphne h)* dying young was not. Although some parents stated that the child's illness was never discussed with and/or in front of the child, it was rarely so uncomplicated. Inevitably there had to be some talk for example, to explain medical procedures or changes in the child's state of health. Participating children and siblings seemed able to illustrate how family members dealt with the subject of the child's life limiting illness. The following quote shows how the sister of *Reuben b)* constructed the complex way that their family talked about the child's life limiting illness even though her parents had said repeatedly in separate interviews *'we don't talk about it (the child's life limiting illness) as a family'.*

> *'My mum will tell you, my dad's lovely and we get comfort off our dad but as far as trying to find out what was going on with my brother we didn't ask dad. My mum puts pressure on herself – No she does, terrible, yes she worries about everybody. Mum is always saying 'don't discuss it in front of Reuben' or 'always talk on the computer' and she is always saying 'Reuben is here' and then she will shut the box because he started reading. No, I mean I know Reuben doesn't know everything … he knows he is a special child and he has got things wrong with his heart and stuff … but I don't think he actually knows the full story but obviously mum explains to him … 'oh you have got to go into hospital and have an operation and put dye in your system' and what have you … so she does explain what's happening but I don't think she actually explains you know … obviously … how dangerous the process is going to be … or …* ***what is going to happen.*** *"*
>
> *Reuben b) - sister (early twenties)*

Knowledge of the child's life limiting illness generated the need amongst family members to talk, but did not provide parents with information for potential questions. Many parents searched for ideas about how to talk to the child and their siblings but found little practical advice. Parents described and tried to take a clear approach to talk about the illness, especially with the child. Despite the fact that participating parents described two apparently simple and clear-cut approaches for talk within the family about the child's life limiting illness – all participating families talked to the child about the short term consequences of the illness and related experiences but none of the families talked to the children about the life limiting nature of the illness.

6.2 Family talk: the child dying

Although none of the families talked to the child about their shortened life expectancy, they made separate and different decisions about how to talk to siblings. Parents did talk with siblings, over the age of five, about the fact that the child would die in childhood. In three families *[Reuben b) Jagger e) and Jagger g)]* parents said they were open with siblings that the child would die young because they had felt uninformed about the terminal illness of members of their own family in their youth. In some families parents spoke differently to siblings of different ages and discussed the changing needs of developing siblings over the years of the child's life.

> *'No, I think it is very hard to sit down and say to your children this is the way it is. But if you talk around openly we find … if they want to ask something … they'll ask me. It's easier for them because they've already heard it and it seems to work. I don't know if it is the right way or not but it seems to work. My teenage son is different now he keeps everything in and doesn't really talk. I think, as he has got older he has got aware of life and death. What has happened as well, although he has always known Daphne has got problems; I think when she was in hospital last time it really brought it home to him. With our youngest it wasn't quite that bad because she was with me, she was with me all the time so she saw everything what was going on."*
>
> *Daphne h) - mother*

These parents were constantly evaluating and re-evaluating the needs of their siblings, especially the changing needs of their adolescent son. In their descriptions they linked their son's changing needs, to his age and experience of a recent critical episode from which his sister almost died, this can be linked to the concept of 'moments of realisation'. The father went on to express his fears about how his son will cope when his sister dies.

> *'I kept saying to him we have to go to the hospital again tonight mate and it's like 'well I really want to be with my friends'. And then when you get home and say 'she is very ill today' he is feeling guilty because he is thinking that might be the last day, he is no different to other teenagers. Like, I put myself in his place and think I would have been thinking I really want to be out tonight. I find that children aged six or less just think 'she is always going to be here', even though you say 'Daphne might die tomorrow' it's like it doesn't matter, it doesn't even stay in our youngest one's head but when you say this to a teenager, wouldn't you say, (asking his wife), it*

> *means something. That's the way I was looking at it, because I was watching them to see their reactions, I was watching my kids and thinking are they going to get over this? Is my son going to get over this?"*
>
> *Daphne h) – father*

Although talk about the child's illness was not linked to any particular conceptual cases there were links to 'moments of realisation' of unfolding life stories *['A cycle of crisis and survival', 'acute loss of abilities', 'life threatening surgery' and moments when 'slow deterioration' were suddenly revealed]*. At these times the threat to the child's life was clarified and this raised awareness and created anxiety amongst family members especially about communication.

Siblings had questions from their own perspective that they did not necessarily ask parents until they were participating in this research. The questions raised by siblings were not always easy to anticipate, making it essential to hear from participating brothers and sisters directly. To illustrate, the sister of *Jagger e)* would not talk about his death until our final meeting. In her mother's presence she explained that she worried about where her brother would die, explaining that at one time she had hoped he would die at home, but later she thought it would be better if *Jagger e)* died at the hospice. She talked about the 'special bedrooms' at the children's hospice and asked a question, which revealed a lack of knowledge despite some information,

> *"Is that 'special bedroom' for children who are dying?" Jagger e) - sister (aged 12)*

I explained that what they call 'the special bedrooms' at the hospice are for children after they die because they can be kept very cool. She did not ask why. We talked about the fact that any bedroom might be used by any child with only a short time to live.

> *"What happens if another child pops into the special bedroom at the same time that Jagger needs it?" Jagger e) - sister (aged 12)*

We talked about the fact that there are two cold bedrooms for children who had died at the participating children's hospice and that a third bedroom could be chilled with an air conditioning unit if it was needed.

On the whole participating parents did talk to siblings about the life limiting nature of the child's illness, although they did not discuss this with the child. The concept of 'moments of realisation' can be linked to times when awareness of the threat to the child's life became more clear, increasing anxiety about communication. Parents worried about the changing needs of developing siblings and siblings (aged two to twenty five) had their own questions about the child's illness and most of these (aged four to twenty five) talked about their own fears for the child's life. Children and siblings outlined family communication 'rules' and described and understood the roles taken on by family members. Siblings talked about the difficulty of talking with the child about their illness whilst maintaining the essence of their sibling relationship but no siblings raised any issues related to their knowing what the child did not know: that the illness was life limiting.

6.3 Child's questions: death in childhood

Children taking part in the research who could communicate verbally and who were cognitively able, raised the subject of death in childhood. Parents or siblings did not raise this subject. The four participating children who could communicate verbally *[Reuben a), Reuben b) Imogen c) and Daohne h)]* talked about their illness on our second or third meeting together; two of the four talked about their personal need to talk about their illness *[Imogen c) and Daphne h)]*; three of the four talked about death in childhood *[Reuben b), Imogen c) and Daphne h)]*; and one of the four talked about her own dying *[Daphne h]*. Children were aware that sometimes adults did not talk about their illness in order to protect them and in a reciprocal cycle the young people would then rarely raise the subject either. The next quote reveals how the child used a television programme to bring up the subject of death in childhood in our work together. Talking about his favourite soap opera *Reuben b)* suddenly fixed his eyes on mine, more then he did at any time in our discussions, "*Did you see it last week?*' I explained that I didn't always watch the programme he was talking about.

> *'Well one of the men he was going to get married but he said he couldn't cause he is gay ...he only likes boys and couldn't marry a girl! The baby died and they put him in a box in the ground and the dad didn't know what to do so he got the teddy and put that in for him."*
>
> *Toni - 'That sounds sad ... I think I would have been sad ... how did you feel?"*

> *"Yes sort of ... like the box going in the ground ... but he was dead ... so that was OK , not really him ... just his body ... sort of thing."*
>
> *Toni – "Well yes when someone has died it is just the body going in the ground not the person. But the people who love them will miss them and feel sad ... like that poor dad getting the teddy."*
>
> *Reuben smiled at me and I asked if they talked about the programme as a family and he said*
>
> *"No! ... Oh no not that one!"*
>
> *Reflecting back on an earlier conversation I said, "You know you were telling me that if you are unhappy your dad sometimes lets you have something sweet to eat and tells you not to worry? Could you ask him about that programme."*
>
> *"Well dad gives me a little spoon of sugar if I need it ... um delicious! But not like talking about that ... it doesn't matter."*
>
> *Reuben b) – (aged six)*

Reuben b) took my hand, led me to a new spot in the garden and suggested we do some more work on his 'newspaper'. When he got to the box labelled *'When I grow up I will ...'* he said,

> *"I got a list but it's just a list ... I'll leave it blank." - Reuben b) – (aged six)*

It is interesting to note that this is a family in which homosexuality was not a taboo subject and *Reuben b)* had an appropriate for his age understanding about the notion that two men can love each other. However, the subject of death in childhood was taboo in Reuben's family: he understood that his parents would struggle to discuss the subject of a child's death and in a cycle of protection he saved his questions for me. Although three of the four children made some reference to death in childhood, only one related this to herself and she used the metaphor of 'Neverland' drawn from the Peter Pan story.

> *Daphne h) – field notes from visit to family home*
>
> *Daphne created some photographic images using the digital camera and taking photos of the reflection on a window. She told me this is a picture of Peter Pan who is coming to take her to Neverland.*
>
> *"I died!" Daphne explained. With her mother's encouragement she changed this to "I fainted and fell down". Several times she confided that she is going to Neverland soon. Again I asked where that is and she said 'up in the sky' I asked how she will get there and she said 'Peter will take me.' I asked is it scary to think of and she said 'a bit scary in the sky but Peter is my best boy and nice when you get there, to play with the lost boys.' I asked, 'A heavenly kind of place?' and she said 'Heavenly, yes'. I cannot be sure what this really means to her, she is very keen on the Peter Pan film and watches it repeatedly. However she raises the subject in connection with discussions of her own illness and critical moments in her life, as she did here.*

My field notes reveal how I hesitated and felt uncertain when *Daphne h)* seemed to be telling me that she knew she might die young using the Peter Pan story. I was influenced by her developmental delay and I told myself she might not be ready for more questions. Looking back I know that I was not ready to ask further questions. Like *Reuben b)*, *Daphne h)* used a film to raise a difficult subject. Later in our work together the mother of *Daphne h)* raised the idea that her daughter used television and films to *'ask her own questions'* about death and dying and might be using the Peter Pan story to comfort herself with the idea that when we die we are not going to be alone. Although the family was not certain that *Daphne h)* was using the Peter Pan metaphor to talk about her own dying the subject was left open. This uncertainty can be linked to 'moments of realisation' – *[The child's life and death]*. Although the young person seemed to be asking about her own dying, uncertainty, fear and the overall focus on life and living hindered the adult response.

Only participating children raised the subject of death in childhood. They used hints, oblique references and metaphors to learn more about topics that were not openly discussed within the family but they were dependent on the reactions and responses of adults for the subject to become more open.

6.4 Experiences like and different to peers

In initial meetings the children described everyday experiences, common to all children. For a time I considered the idea that the children were giving accounts of themselves as 'normal' (like their peers). As the children and I spent more time together they would introduce one or two experiences related to their illness for example a recent hospital visit or respite break at the hospice. Gradually, the children used their time with me to explore experiences related to their illness more and more. The children would move on to exploring differences between themselves and their peers and slip into using medical language and revealing their understanding. The children seemed to use the process of the research to highlight to me, they had experiences that were 'ordinary' (typical of the social groups that they belonged to) before exploring experiences related to their life limiting illness, which they knew were unusual. Children and families focused day-to-day on living but were aware of the ever-present threat to the child's life, which can again be connected to the notion of 'moments of realisation' – *[The child's life and death]*.

Families of children less able to represent themselves would also begin with stories about their efforts to ensure that their child or children had 'ordinary experiences'. Parents and siblings in these families would also emphasise endearing aspects of the child's personality like playfulness, humour or tenacity but the change in emphasis to the consequences and experiences derived from the child's life limiting illness came more quickly than it did with children who were representing themselves.

Parents prepared young children with simple answers to potential questions from peers about their illness or its consequences - for example, scars and decreasing mobility or the inability to participate in day-to-day activities like sports. They also sought to protect young children from some of the themes between the lines of my own questions. Below is an illustration of how a mother interjected the simple answers she gave her child to answer any questions about the scars on his body, before he had time to explore how children in his school knew things about him that he had not told them. In turn he responded to the comfort and obvious familiarity of their rehearsal.

> *Toni - Do the children at your school know you have a new liver Rueben?*
>
> *I think! [Nodding]*

> *Toni – You think they do? Did you tell them?*
>
> *Reuben - No*
>
> *Mum - That's why I told you to tell people it's your new liver didn't I darling…so if anyone at school saw your scar while you were getting changed for PE, so you could tell them what it was…is that right?*
>
> *Reuben - Yeah! [smiling]*
>
> *Mum – Do you remember when we went on holiday and you went in the swimming pool…and I said to you there's that little girl looking at your scar? Do you remember…? What did I say to you to tell her?*
>
> *It's my new liver!*
>
> *Reuben a) – (aged six)*

Simple explanations were most useful to younger children, older children or children talking to adults were managing much more complex systems of talk, questions or more notably silences. The primary role of the child's experiences at school was to reveal to them that they were different to their classmates. *Imogen c)* described how events outside her home clarified to her that she was different from the person she was before her illness and different from the other young people in her peer group. Information about the impact of their illness did not just come to children in the words of other children or adults but through their experiences and personal interpretations of events.

> *'I love school and my favourite subject is maths. I suppose there is one thing … as I said I used to be an athlete. You know! I was taking part in competitions and winning gold medals and trophies. Well I still like PE at school but I think the reason I don't like it as much is that I can't do as much as I used to. I can do stretches and there is this ball with a jingle it … so just mess about with that really. Sometimes I get really frustrated and my teacher says well just watch the others then. I do like school I just get a bit frustrated at PE that's all! Sometimes I think my friends get frustrated cause they think … as well as myself … why do I have to suffer?"*
>
> *Imogen c) – (aged 14)*

Adolescents had greater self-awareness, greater awareness about the underlying motivations of others, interacted with more people and had more opportunities for experiences away from their family home. Parents of adolescents made no efforts to control knowledge and talk about the child's illness amongst their school peers and friends. The young person's peers were sometimes a great source of support and encouragement but the young person's parents had concerns about the emotional impact on other young people.

> *'Well most of them (school friends) have known Imogen for many years … they didn't suddenly meet a child with disabilities. They always say to her 'we want you with us … look after yourself and listen to the doctors!' It is encouraging for her that they love her. But then again, one time I went to the school because she was ill and all the class were in tears … I thought that was awful."*
>
> *Imogen c) – mother*

Children of different ages taking part in the research, who could articulate their thoughts seemed to have a developing sense that they were different to their peers. Statements by *Reuben a)* were confined to relatively simple differences in his overall appearance, for example,

> *Toni's field notes from a session with Reuben a) at his home*
>
> *Drawing a friend in the middle of a sheet of A4 he added himself about a third of the size of the first figure in the bottom right of the page. When I commented 'She looks tall' … he said, 'I bees small'. Reuben a) – (aged six)*

Reuben b) hinted at his awareness of differences between himself and his peers for example after talking at length about being part of the class and providing a long list of *'friends'* he announced *'it's just kids at school they are all different to me.'* The adolescent *[Imogen c)]* was certain about particular differences between her own physical abilities and those of her classmates and friends. However I sensed that *Reuben b)* was more aware of his own declining strength, perhaps because *Imogen c)*'s state of health had remained somewhat unchanged over the previous six months whereas *Reuben b)* had suffered two critical episodes during the same period.

The concept of being different to peers was discussed in some families in terms of the child's disabilities but not in terms of their illness being life limiting or even in terms of their physical frailty or vulnerability. The children themselves gave priority to describing their ordinary experiences in the first encounter with me but it soon became evident that they were aware that many of their experiences were different to their peers, and that these experiences were an indication that they were different. Children of various ages explored 'differences' between themselves at different levels – young children mostly explored differences in appearance but adolescents explored more complex differences.

6.5 Children's hospice experiences

Hospice was the one setting where parents and one young person struggled with social meanings of the word 'hospice': the idea that hospice is a place where people go to die. The referral criteria of the participating children's hospice meant acceptance that the child was life limited, which was discussed openly with parents but not with children or young people. Participating families actively using the hospice services valued the support provided *[Reuben a), Reuben b), Imogen c), Imogen d), Jagger g), Leonora i), Daphne h), Marcus k) and Marcus j)]* but some *[Reuben b), Imogen c) and Jagger g)]* struggled with social notions that using a hospice meant the child was dying or that the family was in some way losing hope by accepting referral to a hospice.

> *'Well I hate the word 'hospice' and even a friend said, 'Why are you taking him there, he is not dying is he?' But the point is it is just great for all of us ... just the help and support we need to keep going. But it's the word people don't understand and I say well no just come and see."*
>
> *Reuben b) - mother*

Children and families recast the meaning of hospice in terms of their personal experiences of respite and support. *Imogen c)* wanted me to understand her initial fears about visiting the hospice but how she had found a new meaning drawn from her experiences of using the service.

> *"Yeah! I heard it was like a hospice ... and I wasn't really keen on going, cause I thought hospices were places where people die ... First of all I talked to my mum ... then I remember*

> *two of the care team came to my house to talk to me. They spoke to me and explained it's like respite."*
>
> *Toni - "Respite?"*
>
> *"Yeah! Like a break for me and my family ... that's all! When I went there it was excellent. There were also two celebrities there that day! And ... it was just excellent ... the way they (the hospice staff) treated us ... they were mad (laughing)!"*
>
> *Toni – "Did you meet other children or just celebrities?"*
>
> *(Both laughing) "I did meet a cute little boy and a girl who is a year younger then me. We had a lot in common and she said she is going to marry David Beckham and I said 'no me!' She is in a wheelchair like myself ..."*
>
> *Toni – "I see and did she tell you what was wrong with her?"*
>
> *"We didn't talk about that ..."*
>
> *Toni – "So when you were worried about going to the hospice and you were told it was 'respite' did you know what that meant really?"*
>
> *"Not really, no! I just tried to make a joke out of it. Like a place where you go and rest and they bite you! (Grinning). But no now I have been on my own to stay and it is just a place where you go and have fun, relax and give your parents a break."*
>
> *Imogen c) – (aged 14)*

Siblings, like the children, focused on the whole on fun and activities at the hospice. In the following example one sibling told me how he tried to offer his support to another sibling visiting the children's hospice. This illustration also underscored how much children and siblings needed to articulate their fears and worries alongside their ability to recognise and respond to the needs of other children and young people.

> *"You have to talk because the other person might have a way to help and you have to get it out ... I don't want to keep it in ... But like sometimes children with special needs get dropped off at*

> *the hospice by their parents and there is nobody else there only other people … no family! Other people bring the whole family … I talked to one boy last time we was there and he was really upset because his brother was very ill. I like talking to other people with the same problem."*
>
> *Reuben a) – brother (aged 12)*
>
> *Reuben a) – brother (aged12) Toni's field notes from visit to the family home*
>
> *Following on from this conversation we discussed how talking about painful emotions does help but Reuben's brother explained that you can't just talk to anyone. In his experience talking to friends needed a feeling of trust; that the other person would be confidential and respect that you might just want them to hear your worries but not necessarily take any actions on your behalf. He told me that when you know you can really talk to someone, that indicates they are likely to be 'very good people … people you could have a joke with as well!'*

Hospice was the one social setting linked to ideas about death and dying. Participants created new meanings based on their personal experiences of using hospice services. They used these to explain to 'others' why the hospice met their needs and the needs of their child who was enjoying life – although their life would be short. This focus on living can again be related to 'moments of realisation' – [*The child's life and death*]. It was important to all of the participating families that the children's hospice took the lead from parents about how to talk with the child about their life limiting illness. This was primarily because none of the families talked to the children about dying young. Children and siblings met other children and siblings at the hospice with experiences related to the child's life limiting illness; they shared experiences that they could not have explored with peers at school. Some took the opportunity to talk others chose not to.

6.6 Parents and death of other children

Three mothers and one father talked about the death of other children in care settings and how the experience of witnessing the grief of other parents highlighted and clarified their emotional fears about their own bereavement

> *"One child was in PICU at the same time as Reuben and he didn't come out. They couldn't get him off the ventilator, he had a liver transplant, liver was working fine no problem but it was the*

> *bloody ventilator, it was his lungs that gave out and it was just like Sod's law you know he had gone through the liver transplant fine and it was his bloody lungs.*
>
> *When he died he was taken obviously down to the morgue and they (the child's parents) went home and I actually said I don't think I can do it, I really don't think I could do it, I said, I don't think I could leave this hospital without Reuben. That sort of gave me a sort of an insight to like how you would cling on.*
>
> *It's like how do you ever know that you picked the right time, how do you not sit there and think what if I had made another decision. Would he be at school now? Would he be laughing? Would he be playing? Would he be you know … how do you ever know it is the right time? As I say I really do think now, I would cling on to all of my children."*
>
> *Reuben a) – mother*

Overall the subject of their own child dying was almost too painful for many parents to articulate. However, they found the courage to talk about death of other children. This seemed to facilitate their own reflections about when their child would die and talk with me or other families.

> *"Yeah well all my kids have lived a life. I'm not obsessed with dying but I will talk about it and I've been able to talk about it with people about their children dying without it becoming too much of an issue for families. No in fact I talked to one lady about her son dying before he got bad and we talked about things that she'd never talked to anyone about and I think that helped her for when he actually did die."*
>
> *Jagger g) - mother*

Although none of the children or siblings talked about this particular experience, clearly they could also get to know other children in hospital or at the children's hospice who might die. Although being witness to the death of other children was distressing to parents it did give them a way to begin talking and exploring their own emotions in relation to the fact that their child would also die in childhood. In addition it gave parents the courage to raise this subject with me and with other parents in the same situation.

6.7 Feeling 'professionally' unheard

Families constructed the idea that professionals communicated differently with each other than they did with families. Like other parents who participated in the research the mother of *Jagger e)* talked of feeling unintelligible, *"On the medical side … it's as though they don't understand what I say."* Feeling unable to make her fears heard, the mother of *Jagger e)* refused to bring her son home from the hospital but expressed discomfort, frustration and a sense of finding no other way to make her fears understood. The parents of *Imogen c)* used the power of language to force professionals to respond to their fears.

> *"We took her to our local casualty and they said she was not well but they didn't have a bed. I had to bring her home. You know she got so lean you could see her heart beating like those starving children you see on television … after a couple of days I took her back to the hospital …I said to them if you were to come in my house and find my child like this you will accuse me of child abuse, do something!"*
>
> *Imogen c) - mother*

Participating parents expressed the idea that professionals often heard each other better than they heard parents.

Describing why he had confidence in the specialist centre, the father of *Imogen d)* drew out the idea that *'negative news'* can be given to parents in positive ways. Despite the fact that the doctors at the specialist centre were still unsure of the diagnosis for *Imogen d)* they were telling her parents what medical conditions they had ruled out and what other conditions they were testing for. The parents of *Imogen d)* felt that at the specialist centre her condition was responded to urgently and systematically and they were communicated with openly.

> *"Now I make a point of telling staff at the local hospital that she is a patient of Dr 'X' from the specialist hospital. Like I know it probably sounds ridiculous. But it is the only way I can get peace of mind. It's surprising how they listen now we have learned everything from the specialist hospital and all the prominent doctors you meet up there. It is really surprising how they listen when you start quoting professors from that hospital. I'd say it is a bit like living in the army and saying you've got a friend who is a General!*

> *It makes them take you seriously, rather than just assuming that you are some council estate urbo!"*
>
> *Imogen d) - father*

Experience had taught the father of *Imogen d)* that he would remain unheard unless he spoke the *'right language'*. He had learned to give a clear account of his daughter's condition and medical history and he made overt references to links with the *'specialist centre'* and *'prominent doctors'* in order to reposition his child and himself in the hierarchical world of the local hospital: *'like living in the army and saying your friend is a general.'*

It was implicit in much of what families said about interactions with professionals that professionals rarely communicated directly with the child about their illness. One child drew out the notion that he was not meant to hear what professionals said to each other about him in respite centres and at the hospital. Having made this revelation he would provide no further detail and left me wondering how he actually perceived my role and place in the children's hospice, where we were talking, or did he suddenly wonder whether we might have been overheard? Only speculation - I learned that sometimes children taking part in research only reveal glimmers of ideas and leave you uncertain of their full meaning.

> *Reuben b) – field notes from meeting at the children's hospice*
>
> *He provided a long list of favourite care staff. I listened and then asked where else he went for care. He described a charity funded respite centre and so I asked what it was like to visit. He said, 'Oh it's a big house like this one ...but smaller with a drive. They don't look after you, your mum does it.' He thought for a while and then said, 'Well they look after all of you then you just be together but it is not so fun as here (the children's hospice). He explained further, 'When they come to look after you they have 'handover' out there so you don't listen.' I asked what 'handover' is and he said, 'when the nurses tell each other how you are and who is going to look after you and all that.' I asked why he couldn't listen, he said he would talk to me later and ran off to find out who his care team member was for the afternoon.*

Individual family members were critical of the number of professionals involved in their lives but greatly valued individuals who communicated with the whole family and recognised the needs of each person.

> *'Basically when my brother is ill a lot of people get involved: you've got the family doctor, the hospital, the pharmacy, the ward staff, the hospital care team in casualty and the paramedics.*
>
> *Then the other side of it you've got the respite people and carers. Like at the respite centre there are two ladies there called Mary and Jean and we have known them so long we call them aunty ...like part of the family.*
>
> *Then you've got the carers who come here ... like I call one of them the Thursday carer.*
>
> *Then there are carers from the hospice who come now and again ... say when a nice movie comes out and I want to go and see it then I can't see it ... one ... because we can't get Jagger in the cinema and ... two ... he would be too noisy and we would get chucked out. So it's like nice ... when one of them come to look after Jagger so that we could go out.*
>
> *Rebbeca was the very first person that came to us I think and I do miss her! Yeah (laughs) but she done a lot for the whole family if you see what I mean ... like ... me, mum, and Jagger! Jagger got caring. Mum got time to go out or have rests, and I got like someone as a friend really. After when you get used to it you know the different carers come and go."*
>
> *Jagger e) – sister (aged 14)*

Participants were sceptical about the ability of professionals to understand their family life partly because of fleeting relationships combined with the fact that at many of their worst times, they were alone with the child.

> *'Professionals say they want to understand what it's like day to day but carers breeze in ... they breeze out. I don't mean that the way it sounds ... But they are not here when the bottom line is that Jagger's fitting really bad or going blue. Not here when we have to hit the nitty gritty where poo goes everywhere and that's the nitty gritty stuff about caring! As soon as you are a parent it's guilt and as a parent you got to have courage ... right? All parents! With Jagger it's been learning when I can keep looking after him at home and when we need the hospital. Where I've*

> *learned to wait that bit longer ... you might be more borderline panicky! But if I get it wrong ... then the guilt."*
>
> *Jagger g) – mother*

Many parents talked about the difficulties of funding. Getting required equipment was through a slow and difficult process for children like *Leonora i).* Funding and resources were complicated and limited in terms of transitional care for young people like *Daphne h).* Many parents put forward the idea that, although professionals working within one health or social care setting seemed to hear each other better than they heard families, 'agencies' failed to talk to each other, especially over funding issues, which caused delays in meeting the needs of children with limited time to live.

> *'They have these meetings and it is really difficult for parents. They have multidisciplinary meetings and you must have been to these, social worker there, nurse there, consultant there, me there, mum there, another nurse, some health visitor there, you have got a speech language therapist, someone from social services and anyway – the whole lot and you go round the table and explain who you are and you just take part in this and they just treat you as if you don't exist, it is awful there is two of you and they --- gang up on you! Then you have to try and deal with all these people who are not all that helpful who don't talk to each other. At the end of it ... all they agree on is to have another meeting!*
>
> *Leonora i) – father*

Overall participants complained that professionals listened to each other but not to families and they rarely spoke to the child about their life limiting illness directly. One father put forward the idea that professionals can give negative news to families in positive ways. Other participants were critical of the numbers of professionals involved in their lives and the paradoxical lack of practical support combined with delays over funding issues.

6.8 Social reactions and judgements

Many of the children, most of their siblings and some parents talked at length about social reactions to disability not just in relation to their own experiences but also in their observation of other children - for example, at school and the disabled children of neighbours. The mother of *Jagger f)* expressed the idea that 'disability' was still a subject that was a social taboo *"people don't like to talk about disability but this problem is out there isn't it?"* The sister of *Jagger e)* felt that 'social staring' at disabled people was associated with wider judgements not just about the person and their right to life but also about families of disabled children. She was critical of society's inability to see past the obvious *'a wheelchair'* to the individual qualities and needs of each person. In a comment repeatedly mirrored by other participants, the mother of *Jagger g)* suggested that children have a more natural or acceptable way of dealing with disabled people … they ask questions. *Daphne h)* overheard me reading a book called *'I'm special'* with one of her siblings and specifically asked me to bring it for her next session. Turning straight to the pages illustrating how sometimes people stare at disabled children, *Daphne h)* told me, *'I don't like people staring at me it's not lady like I am an adult!'*

> *"Not many people are like my family … there's a young girl round the corner and her mum don't take her out. I think it's because she's disabled and her mum can't handle that people are going to stare. Even we get wound up a bit! It's just like the funny looks that they give Jagger like really horrible looks … as if to say, 'What on earth are you doing on this earth you shouldn't be here like that kind of thing. Yeah judging (pause) like judging other people all around basically because they're a different colour, because they're disabled, because of their religion, so all around kind of thing, that's what I don't like.*
>
> *Toni - Do you think people judge Jagger because of his problems?*
>
> *Umm. I think they judge like more … me, my mum, my little brother and Jagger! Like … 'oh my God they've got that in their family'… 'oh my God! … errr! None of them should be on this world and things like that. That's what I think they're thinking basically the way that they give the looks and that. I think with a disabled person the first thing people see is the wheelchair. But everyone is different … some of them are disabled because they can't walk …or they can't speak up and they've got like trouble learning …but the end of the day we are all different."*
> *Jagger e) - sister (12)*

Participants hardly mentioned any social opportunities to talk about the life limiting nature of the child's illness. Their omission indicated to me that the subject was rarely disclosed. The parents of *Reuben a) and Reuben b)* did highlight poor social understanding about their son's illness. Referring back to the conceptual cases *'Reuben'*, these were the only participating families who felt judged because their child looked 'normal'.

> *"I think people look at Reuben and I almost feel they are accusing me of like well you said he is life limited but he looks so normal and he is still here! People even say, 'Oh well you are so lucky he is still here and anyway you knew didn't you from the start'. Like it won't be a shock if anything happens to him."*
>
> *Reuben a) - father*
>
> *"I have tried to explain how I feel to make them think. The only way I can explain is ... imagine if you were taking your child to school on a Monday morning and somebody came and said they were going to have your child one Monday. Not tell you what Monday. Just leave you knowing that one Monday you are going to arrive at the school gates and he won't be there. I tell them that is what I feel like everyday ... someone is going to take him one day and I have no control. Imagine living with the pain and frustration of that every day ... of never wanting to leave your child at the school gate."*
>
> *Reuben b) – mother*

Participants described the idea the disability was a social taboo and that adults in society stared but did not communicate with the child and their family. Participants, especially the children and their siblings felt isolated and judged. It was clear that there were few if any, opportunities for children and their families to talk and explore their emotions about the child's life limiting illness.

6.9 Summary

Hearing that their child had a life limiting illness created the need for communication between the child and family members. Participating parents wanted to protect the child and siblings and they feared that talk about the child's illness would raise unanswerable questions. Participants separated the subject of the child's life limiting illness into three topics i) the illness, ii) death in childhood and iii) the child's dying.

Many participating parents talked about the physical consequences of the illness and care and treatment experiences with the child, regardless of the child's ability to comprehend or respond. This communication was given alongside physical care and comforting; it was part of parental care and love to talk to the child and offer explanations. Parents talking to children who were able to consider their own experiences and ask questions faced a different dilemma. Although parents in two families expressly stated that they tried to control how the child's life limiting illness was discussed with and/or in front of the child, in practice none of the participating families talked to the child about the life limiting nature of the illness. Participating parents searched for ideas about how to talk from a range of media and through taking part in this research. Parents worried about how best to evolve the story of the child's illness for the child over the child's life as they grew and developed.

Although parents did not talk to the child about the child's short life expectancy, they did talk to siblings about the child's dying young. Some participants specifically stated that they talked to siblings because they had felt uninformed about the illness or deaths of family members in their childhood. The decision to talk to siblings about the child's dying was different and separate to decisions about how to communicate with the child. The need to talk about the child's dying was heightened at moments when the child was critically ill or when their condition had deteriorated in some way (recurring 'moments of realisation' in the child's unfolding life story). Families shared experiences of visits to hospitals at these times and described increased anxiety about the child's death and the need to talk to siblings. Parents worried about how siblings would cope with and survive the child's death. Siblings had their own questions about the child's illness and death that they asked during the research and which were not easy to anticipate. Participating siblings seemed to need time and space to find the moment to ask and to be heard. No participating families raised any questions about siblings knowing what the child did not know and no participating siblings revealed their knowledge to the child.

Participating children raised the subject of death in childhood. Some children indicated that the subject was taboo in their own family. One young person used metaphor to raise and explore the subject of her own dying but she was faced with adults who doubted her meaning and worried about how to respond without upsetting her. Children were insightful about the emotions and fears of others and were especially protective of family members; they would largely follow the family approach to talk about their illness.

The children and families taking part in this research were actively trying to protect each other from the long term and daily impact of the threat to the child's life from their illness. The children who could talk to me were continuously evaluating their experiences in relation to their sense of themselves and this influenced how the children represented themselves within the research, which changed as we spent more time together. The children were clearly aware they were different to their peers but in initial meetings with me they emphasised their 'ordinary' experiences. The children did not say *'I am normal'*; they described how they had lots of experiences like the experiences of other children. In later sessions with me, the children began to share experiences that were different to their peers that they could not make sense of, or that challenged their sense of themselves and family strategies for discussing their illness.

Some participants worried about social meanings of the word 'hospice' and links to death and dying. Participants valued the children's hospice service and tended to recast the meaning in terms of their own experiences of respite and fun. Participants felt that the hospice followed the family approach to communication about the life limiting nature of the child's illness especially with the child. This was important to all of the participating families but particularly to parents who wanted to control how the child's illness was discussed with them. Children and siblings also identified the hospice as the one place where they met other children and siblings who had experiences linked to the child's life limiting illness and some used the opportunity to explore experiences that they could not explore with peers at school. Others adhered to family communication strategies and only referred to the illness in the terms used within their family.

Although the subject of their own child's dying was too painful for most parents, some had witnessed the death of other children in health care settings. Whilst parents found these experiences distressing, they did seem able to explore their own emotions and fears through talk of what they had witnessed. The subject of 'other children dying' was one that parents used to explore their own fears in an indirect way, comparable to the way the children used stories and metaphor as a bridge to the subject of death in childhood.

Many participating families complained that they lived in a context of poor understanding. Participants frequently complained that professionals only listened to each other and agencies failed to communicate when funding was needed. It was suggested that being away from home was often uncomfortable because people in society stared and made judgements but rarely demonstrated any deeper understanding about the child's life limiting illness. Families felt isolated with the knowledge that their child would die young. They felt poorly supported, socially misunderstood and that the subject of life limiting illness was a social taboo. This context further complicated how families talked to the child about their illness because there were few, if any encounters in which families could talk and explore their own emotions or questions about how to talk to the child about their illness.

Chapter Seven

7 Identity, developing self and life story

The aims of this chapter are to explore the impact of having a life limiting condition on a child's developing sense of self, to ask whether the children unable to communicate verbally had active or passive roles in shaping their identities and explore how parents and siblings represented the children who were least able to represent themselves.

The following children were able to actively represent themselves *[Reuben a), Reuben b), Imogen c), Imogen d), and Daphne h)]* through interaction with me, verbal language and British Sign Language. Other children communicated non-verbally in subtle ways that were difficult to interpret in just a few meetings *[Jagger e), Jagger f), Jagger g), Leonora i), Marcus j), and Marcus K)]* and they were often represented by their parents and siblings.

7.1 Integrated self and identity

In some cases the life limiting illness was diagnosed early in the child's life *[Reuben a), Reuben b), Jagger f), Daphne h), Leonora i) and Marcus j)]*. In other cases the life limiting illness emerged early although the underlying medical condition was never diagnosed *[Jagger g) and Marcus k)]*. What these cases had in common was that the children and their families lived with the consequences of the life limiting illness throughout the children's lives. The child's life limiting illness was revealed in their body, through social interactions and in their unfolding life story and seemed almost inseparable from the child's identity.

7.1.1 Integrated physical signs of the illness

In all cases the child's life limiting illness was revealed in their physical body in some way. Signs and symptoms included - for example; jaundice, cyanosis, scars from surgery, curved spines and misshapen limbs. The young children who were able to actively interact with me showed me scars and talked about being *'blue'* or *'small' [Reuben a) and Reuben b)]* but they rarely entered into any detailed comparison of physical differences between themselves and other children. Siblings would sometimes mention physical signs of the child's illness that they were not going to add to artwork about their brother or sister. Signs of the child's life limiting illness were mentioned by siblings but rarely discussed in any further detail.

> *'He doesn't like to talk about being yellow though … I am not going to put that on my spider chart!' - Reuben a) - brother*

In the case of *Reuben a)* there was a strong theme within the family about him being small and all family members reflected on this notion with me separately. Looking at some family photographs together *Reuben a)* commented about his size using humour,

> *Toni's field notes from a visit to Reuben a) at home*
>
> *In one photograph Reuben was standing in the bath whilst his sister was sitting next to him. He grinned at me and said 'I am bigger than my sister there!' So I admired how much bigger he looked and he said, 'Only cause she couldn't get up but I could!*

Although this seemed a simple statement of fact, in that he was a little smaller than other children of his age, in the context of his life story it became clear that for his parents there were deep-rooted links between the idea that he would be very small and his life limiting illness. His father's retelling of the moment he was told about Reuben's diagnosis illustrated this:

> *"Well going back to the diagnosis. They said 'syndrome' and we both jumped years ahead to like what did that mean about his life and that? After another little while there was a lady - came to see us who is the clinical nurses specialist for liver disease. Absolutely brilliant lady, knows her stuff, very reassuring, so we sit down with her and she says, 'Well on this leaflet it says about learning disabilities, well it is not really learning difficulties, instead of being top of the class, he is liable to be in the bottom half of the class as opposed to a fully fledged learning disability where they can't cope at all. Right, the other thing it says on here is dwarfism' and I thought, 'Oh flaming hell', she said, 'Well that is not strictly true either, he will just be a bit shorter than the average.' And I thought, 'Oh okay then'. Um,… I think the, am pretty sure growth retardation was the term used and she said well most people think of that as being dwarfism and I thought 'flaming hell!' …" Reuben a) father*

Families representing children less able to represent themselves, talked more openly about physical differences between the child and other children than families where the child was actively participating in the research. However, all families generally played down physical differences that the child was born with. This was illustrated in the conceptual case *'Jagger'* where his mother talked about his feeding problems but linked them to similar problems experienced by 'many other children'. The mother of *Jagger f)* described how he did not breastfeed *'like lots of other babies'.* He had to be tube fed, *'like lots of other premature babies'* and only when his weight loss was linked to a significant deterioration in his general condition did his mother highlight the change in the body *Jagger f)* as an important sign of his life limiting illness. It was as though physical differences between the child and the average child of a similar age were obvious and could therefore be talked about openly. However, families demoted visible differences in their efforts to help me meet the unique person within, unless or until physical changes demonstrated acute or gradual deterioration in the child's state of health.

The idea that the child was born small (prematurely in many cases) or had feeding difficulties that were difficult to resolve without medical intervention - for example, via naso-gastric or gastrostomy tube feeding - were common across cases where the life limiting illness emerged early in the child's life. Parents often linked the child's general vulnerability to their size and complex nutritional and feeding needs.

Where a life limiting condition emerged early in a child's life, physical signs of the illness were integrated into the child's self and identity. A sense of the child as a person was bound up with basic physical signs of their life limiting illness present from the moment the child was born. This was evident from children actively representing themselves and from parents and siblings representing children less able to represent themselves.

7.1.2 Integrated social identities

The children taking part in this research had many social roles and identities just as we all do: they were members of a family, they were school children with classmates and they were frequent visitors to health and social care settings where they became well known.

Some children used their social interactions to know themselves and to help me know them better *[Reuben a), Reuben b) and Daphne h)].* Accounts of social encounters brought forward notions of the child having experiences 'like other children' and having experiences 'different to other children' but experiences related to their life limiting illness were a part of their whole lives, not something new or separate. In their accounts the children used ordinary social encounters as a way to locate themselves within a 'normal childhood' but knowing they had experiences that other children did not (illustrated through this study through the idea of recurring 'moments of realisation' of the child's life story, combined with repeated hospital visits and visits to the children's hospice).

I found it more complex to get to know some of the children in this group who communicated non-verbally *[Jagger f), Jagger g) Leonora i) Marcus j) and Marcus k)]*. To illustrate, *Jagger f)* communicated with eye contact, smiles and by getting physically close to you. On the other hand, *Marcus j)* would laugh during play or look away if he was bored with your presence. Although I could see that *Leonora i)* took an active role in social interactions, at some levels it was less obvious to me than the active participation of *Jagger f)* or *Marcus j)*. I found this difficult to comprehend until I was talking to her mother, who began to describe *Leonora i)* as a person. Then I found that my field notes illustrated how I initially responded to *Leonora i)* without seeing the individual beyond the physical presence of a child I did not know but wanted to comfort.

> *Toni's field notes from participant observation at home with Leonora i)*
>
> *"I wanted to hold Leonora but was not sure how to do it. Her community care team member from the hospice showed me. I cuddled Leonora for a while and I found her very heavy and difficult to hold because she was tall and had long limbs, which would go into spasm. She was able to hold her head up at times but not at others. I found that she sat best with her legs folded or bent at the knees. I found it hard work to sit on the floor unsupported and comfort Leonora."*

My account illustrates my own awkwardness in trying to meet the needs of a child physically disabled by a life limiting condition but tells the reader nothing about *Leonora i)* as a person. After her mother talked about her personality and identity, I found a short note in my records that provided a much better introduction to *Leonora i)*:

> *Toni's field notes from a visit to Leonora i) at her home*
>
> *Leonora was laying on a wedge in the front room of her home and watching a disco ball. Leonora watched the light quite intently. She vocalised from time to time and moved her limbs and head. I spoke to her and she made eye contact and smiled. Her mother said, 'Oh you are honoured she doesn't normally smile at new people'."*

Families of children unable to communicate verbally who had lived with their life limiting condition all their lives, presented information to me about the child's social encounters and reactions on their behalf. I realised that the child's unique identity was most clear in accounts that made some reference to the child's personal reactions and responses to other people, no matter how subtle.

All of these children lived their whole lives with experiences linked to their life limiting conditions – for example visits to the hospice – and with intrusions from the illness into their daily social roles – for example, they were frequently away from school for routine or emergency hospital visits. Whether the child was able to represent their own social roles or families presented these on the child's behalf, being born with a life limiting illness influenced the child's social interactions throughout their lives.

7.1.3 Integrated life stories

Children born with life limiting conditions were 'a life limited child' from that moment forward. It seemed that for the children who could tell me, their illness was integral to their lives *[Reuben a), Reuben b), Daphne h)]*.

> *"I've been to the hospital hundreds of times. Sometimes, emergency sometimes just going."*
>
> *Reuben a)*
>
> *"Draw about being me! I know I'll draw me in bed … but I'm not sick and I'm not in hospital!"*
>
> *Reuben b)*

One adolescent with learning difficulties *[Daphne h)]* described how she collapsed and was rushed to hospital in an ambulance with the idea that she *'died'* and that ambulances are *'scary'* whilst hospitals are *'boring'*. Although she made reference to the notion that she could still *'die'* or *'faint and fall down'* (as she reverted to her mother's words) overall she talked about her life as if she had a future and was longing for some sense of independence. Somehow she was able to maintain aspirations for the future, alongside the idea that she had critical episodes of her illness and could die: an incongruous dichotomy that she seemed able to view in tandem within the context of her own life story.

> *"I am seventeen and a student. I have a boyfriend at school and we hold hands but not kissing that is disgusting! When my ulcers start I get bad pains. I died … I get dizzy, people shout and I fall down. I dance in my wheelchair. I am getting more of a life soon. I'll get married and have kids. If my kids answer me back I will say, 'Stop it that's rude' and if they are sick I will take them to the doctor."*
>
> *Daphne h)*

The children who could, asked questions about their own life story in a rounded way and were just as likely to ask for details of fun family memories, as they were to ask about their illness. When families were willing to explore the child's life story with them, the relaxed exploration seemed to facilitate openness to the child's questions. The children in turn seemed fascinated by stories of their own life and introduced questions about their illness in the context of their whole life. I witnessed a young child *[Reuben a)]* becoming absorbed by his mother's stories of his life, listening intently and seeking more information but also changing the subject and wandering off when he seemed to have heard enough but returning to the subject in his own time.

Children unable to actively participate in the research were represented similarly by parents and siblings *[Jagger f), Jagger g), Leonora i), Marcus j) and Marcus K)]*. Although the child's illness changed the life stories of family members, it was inevitably part of who the child was.

> *"No, but I mean you cry for two weeks and then you have to get on with it because at the end of the day he is just himself! He is a precious loving little boy aren't you Jagger?" – Jagger f) – mother*
>
> *"Leonora is a very resilient child! The number of times she has been at death's door and refused to give up. She wants something! She has her own wish for life." – Leonora i) – father*
>
> *"Marcus … he was always a pipe and slippers man! (Laughs) Basically if you are going to cuddle him and read a story then he is happy to sit there all day, he has always been like that. Now he has deteriorated and that … but I think there is too much giving up and letting him just*

> *sit there all day ... people forget he has always been lazy ... that is who he is ... it is not time to give up on him yet." Marcus k) - father*

The effort of telling the child's story did not provide these families with opportunities for communication with the child. The child's life story was presented as a chronological précis over time with me. Families caring for children unable to communicate verbally emphasised the child's unique qualities, endearing behaviours and traits, social experiences and range of social roles.

In the following cases *Reuben a), Reuben b) Jagger f), Jagger g), Daphne h), Leonora i), Marcus j) and Marcus k)* the child's identity was bound to their embodied presence, social relationships and whole life story and their life limiting illness was always an integral part of their life story. There seemed to be a sense of continuity to the way that these children represented themselves and the way they were presented by family members.

7.2 Loss of past self

In some cases the life limiting illness emerged later in the child's life, even if only two or three years after birth *[Imogen c), Imogen d) and Jagger e).* Although the illness and the threat to these children's lives were clear from the outset of the illness and thereafter, none of these children had a definite medical diagnosis at the time of the research. Only *Imogen c)* was able to talk to me, *Imogen d)* was beginning to learn sign language but had a limited vocabulary and *Jagger e)* communicated with gaze, eye contact and subtle changes in facial expression. The families of *Imogen c), Imogen d) and Jagger e)* had begun to know and then lost the child who was developing before the onset of the life limiting illness.

7.2.1 Changes to the child's body

Families of the following three children, *Imogen c), Imogen d) and Jagger e)* began their stories with the child they were getting to know, a child altered by the onset of symptoms often unusual or unusually severe and beyond the immediate control of physicians. Parents had clear memories of the child who was and they frequently used photographs to show me the child captured visually at some point in time before the onset of the illness. This is captured in the conceptual case 'Jagger'.

> *"No she was a beautiful child. Everyone looks at that photo and says that. She even looks different now. Like looking at a different child. The doctor was here one day and she said, 'That is what you have lost ... (pointing to the photo on the wall) that child in that photo'. It made us cry but it is true."*
>
> *Imogen d) – mother (father nodding)*
>
> *"That's her before the illness (a family photograph of the child) she used to look so cute ... she still looks cute but not so ... strong and well, I guess she has changed a lot! She has looked really thin and then chubby with the illness but never exactly how she used to look"*
>
> *Imogen c) - sister*

It seemed that the joy of sharing these images was clouded in the overwhelming pain of knowing that the child in the picture was gone or had changed profoundly and permanently. The child was no longer the person who was emerging before the illness; they were a new person. Both *Imogen c)* and *Jagger e)* had lost the ability to walk as a consequence of the illness. The mother of *Jagger e)* described how the illness changed his physical abilities

> *"He lost everything he could do before ... everything!" Whilst showing me a family video of Jagger e) toddling, talking and laughing his mother said, "No that's him then! I know he has changed now but I would give anything to see my son walk a few steps again ... anything!"*
>
> *Jagger e) - mother*

It was not until our last session that *Imogen c)* indirectly brought up the issue of survival in a poem that she left on the Dictaphone for me. I asked her if her poem was about the meaning of different people's lives to themselves and other people. She smiled at me and said it was. I asked if she ever worried about her own survival. *Imogen c)* sat quietly for a moment and I worried that I had upset her but I waited (counting to five in my head). She looked thoughtful and then she said,

> *"You know when I couldn't walk ... at that time ... when I first couldn't walk ... I asked the doctor 'Will I ever walk again?' and she said, 'Well we don't know' but that was my worst*

moment, cause I used to be an athlete winning gold medals and all that and I knew then that I had lost all that."

Toni – "Imogen do people ever talk to you about your illness and how serious it is and how much you have lost?"

"They don't you know! Only you and a lady at the hospital who played with me."

Toni – "Does it feel better or worse if people talk and ask questions?"

"Oh much better then you get it out of yourself and it goes away, otherwise it would just sit inside you." (She used her hands to emphasise pushing them away from herself).

Toni – "I wonder why people don't ask?"

"They don't want to upset me … I suppose I don't want to upset them so I just smile and don't say about it either. We all need to get it out sometimes though."

Toni – "So what about you?"

"Oh when I get frustrated I just shout at mum or my sister. Just shout at them and then they give me time to cool down. You know once we were out shopping and my cousin suddenly started to act like a real brat and my mum suddenly goes … 'What is wrong with you?' and he blurted out … 'I'm worried about Imogen because she has been so ill!'"

Toni – "So why was he acting up?"

"Well when you are young, its hard to get it out any other way, maybe you can't say it, so it just pops out like behaving like a brat."

Toni – "I see, what did your mum say to him?"

"Oh just cuddled him and said 'OK you are worried about Imogen … still no need to act like a brat child!" (She giggled putting her hand over her mouth).

Imogen c) – (aged 14)

In general *Imogen c)* made little or no reference to the physical changes brought about by her illness but she took this opportunity to explore changes in her physical abilities and the identity she had before her illness.

Children and siblings who participated in the study were insightful about the emotions of others and especially about the particular difficulties faced by young children who may not have the vocabulary to express their worries and fears. *Imogen c)* used the incident with her young cousin (who had no way to express his sadness about her illness other then to *'act like a brat')* to reveal and explore her own painful emotions. Opportunities for *Imogen c)* to express her frustration at what she had lost only arose within the family but, even within her family, talk about the consequences of her illness was limited. *Imogen c)* made it very clear to me that she preferred to focus on positive aspects of her life but she sometimes needed to communicate about the negative consequences of her illness.

Where the participating children's life limiting illness emerged during childhood they were affected by permanent changes to their physical abilities and general appearance. Some of the children and all of the families had memories of the child before the illness and physical changes were included in presentations of the child.

7.2.2 Assimilated social identities

Some physical changes caused by life limiting illnesses that emerged during childhood impacted on social roles and identities in a variety of ways: two of the children suffered sensory impairment as a result of their illnesses *[Imogen c) and Imogen d)]*. The adolescent worked to assimilate physical changes into her new identity but was aware of overarching changes to her social identity originating from changes to her physical abilities.

Imogen d) eventually lost her hearing and speech, *Imogen c)* lost her sight. *Imogen c)* told me that she could only see changes in patterns of light and she talked at length about facilities and equipment for children with profound visual impairment but she also worked against being labelled as 'blind', arguing that it was not the correct thing to say and participating in activities as though she could see. I commented on this once and her mother said,

> *'If you didn't know her you might not even notice that she cannot see."*
>
> *Imogen c) - mother*

Although *Imogen c)* could describe difference between herself and other young people and had self-awareness about the limits of her abilities, she always worked to be known as a person first and foremost. She was so adept at this that I witnessed many people at the hospice interacting with her for the first time as though she were a child who could see. *Imogen c)* like other adolescents taking part in the research worked to influence how she was perceived by others. She knew that her identity had been changed by the illness but she worked to assimilate changes into her new identity. *Imogen c)* had a range of social relationships in which her primary means of contact and communication was via the telephone. Her sister explained,

> *"On the phone she is equal with her friends; on the phone she can just be herself and her disabilities become invisible."*
>
> *Imogen c) – sister*

The father of *Imogen d)* assimilated changes in his daughter's abilities into her new identity.

> *"She loves other kids, always wants to make friends but she can't chat to them anymore and some get a bit scared. She will point things out to them and try making friends that way … if they look smaller than her she takes on this mother hen role and will ferry them about and be all protective … she wasn't like that so much before but it is cute to watch. She is lovely … different but lovely."*
>
> *Imogen d) - father*

Imogen d) was learning sign language, she could answer and ask simple questions or request her basic needs - for example, she could ask for the toilet or food - but she was unable to explore more abstract topics. Looking at drawings of a disabled child (in the book I'm Special) *Imogen d)* signed, *"My sister"* at the image of the child in a wheelchair left out of the activities of other children. Children were not always able to explore these ideas in any detail and I learned to accept simple statements. On a day out with her from the hospice I witnessed how difficult it was for her to interact with children of her own age.

Changes in social identities brought about by the life limiting illness were described by children, noticed during participant observation or highlighted by siblings and parents.

However, new social roles were assimilated by participants into the child's self and identity.

7.2.3 Assimilated life stories

The onset of life limiting illness in childhood altered the life stories of participating children *[Imogen c), Imogen d) and Jagger e)]*. Only *Imogen c)* might have explored this with me but on the whole she folded the illness into her life story as if it were part of who she had become and the illness seemed inextricable from the story of her life. I got the impression that *Imogen c)* had limited memories of her life before the onset of her illness. Although she did mention changes to her physical abilities, she never dwelled on her losses or what life was like before the illness.

> *Toni's field notes from a home visit to talk to Imogen c) – mother*
>
> *Imogen's mother talked at length about the changes in her daughter's abilities since the onset of the illness, alongside the consistency of her character and spirit. We began to talk about Imogen's school and her mother said, 'Most of her friends have known her since they were at nursery school together. They didn't just meet a child with disabilities who can't see or walk. They watched her through the illness and all the ups and downs since but … she is still Imogen to them! They go shopping together and stay the weekend at each other's houses now and again. They worry about her and look after her, they love her still."*
>
> *"This is who she is and she would be the last to complain, never does! But I look at other kids of her age and I think my sister would be doing that … should be doing that! But she does enjoy her own life but she does miss out on stuff too."*
>
> *Imogen c) – sister*

Parents and siblings representing children less able to represent themselves focused on the child as a unique person living in the moment. The child's life story was told in a chronological format (as if they had told it many times, which I am sure they had) with little or no reference to the immediate or long-term future or to the life the child might have had.

> *"The sweet shop in the train station is a little double-sided kiosk. He's got a shutter that actually opens up on the platform and then there's another one on the other side. Imogen thinks that's the job she really wants! Because she loves trains and there's a man who's got loads and loads of sweets ... as far as she can see - he stands and eats sweets and watches trains! Lovely! The perfect job! (Laughs) No but you can't think about what they might do tomorrow let alone further down the line. We just take one day at a time ... like that's all there is!*
>
> *Imogen d) – father*

> *"Yes ... no this is life now hey Jagger? We do have lovely times and we make the most of them!"*
>
> *Jagger e) – mother*

The child's life limiting illness became a part of their whole life story – it was only barely touched on as something different or new. The children and families did not put forward the notion of the child as a person with a disrupted biography. They had become people with an anticipated short biographies but this was assimilated into their life stories.

7.3 Disruption to the life stories of parents

All participating parents talked about changes to their own life brought about through caring for a child or children affected by life limiting conditions *[Reuben a), Reuben b), Imogen c), Imogen d), Jagger e), Jagger f), Jagger g), Daphne h), Leonora i), Marcus k) and Marcus j)]*. Primarily parents lost or had to make changes to the employment roles that they might otherwise have continued or taken on. This reduced the number of social roles they had and altered the financial status of the family.

> *"I am an arts teacher. I worked full time at first ... then I had to be off when she was sick and they were good about it - but you can't keep being off - not in the current education system. I work three days a week now that seems to work. I suppose I would have stayed full-time but it just didn't work and you have to make a choice ... well not so much a choice, what you need to do for your child."*
>
> *Leonora i) - mother*

In families where one parent worked and the other took on the role of primary carer for the child *[Reuben a), Reuben b), Imogen d), Jagger f), Jagger g) and Marcus k),* some working parents talked about it being easier to go to work.

> *"My husband looks after Imogen and I work. My job is quite stressful but to be perfectly honest I prefer going to work to being at home. I think caring is harder than going to work. And I do. Really! "*
> *Imogen d) - mother*

Some parents talked about feeling they should give up work alongside the guilt of making the decision to stay in employment. Many parents who were working highlighted the pleasures of social interactions through employment.

> *"The hardest part for me was going back to work, I mean obviously we got into a lot of debt. The hardest thing for me was when I did get the full time job having to make a conscious decision that I wouldn't be as close to Leonora as I was in the first six months and to let her go … very difficult decision to make. Um, no real choice … but! It's complicated though because I do enjoy work and everyone is good to me! You work for the money and then you get company and friends who are good to you and if you gave it up for your child you would lose all this too … um!"*
> *Leonora i) – father*

Families where both parents were unemployed expressed some feelings of difference to other families and the idea that they were judged by other families where one or both parents were able to maintain jobs.

> *"A lot of mums at the school don't work, but then their husbands do … again I feel different … sort of … like well neither of us works." Reuben a) – Mother*

The day-to-day impact of the child's illness placed extra demands on couples: they had less time to spend together than they would have had and they did not always agree about family approaches to communication about the child's illness. Whilst trying to make decisions about the best way for family members to talk about the life limiting nature of the child's illness and daily consequences of the illness and/or treatment, parents explained the impact on themselves, their personal needs in terms of communication and the impact on their relationship with their spouse or partner. Couples described missed opportunities for time to themselves and talked about not being able to settle even if they did get a chance to go out with their partner.

One couple struggled to communicate with each other but I realised in their separate interviews that they had quite clear perceptions about each other's needs although they did not seem to know how to support each other. The father became upset during his interview but asked to continue.

> *"It's not necessarily a bad thing you have to confront it. I think about it all the time but I just try to be strong, work hard and keep busy. My wife would say I opt out but it's not just that I don't think it's right to discuss our problems together because it is the same problem and you are just burdening each other. I don't think that is helpful at all, not all the time, it is the same problem and we feel the same way. How can that help? The thing for me also is I don't know how to deal with babies – Leonora and I we could really start to communicate if she gets to the toddler stage, talking and getting interesting. I struggle with the baby stuff. It can be very lonely being a dad. "*
>
> *Leonora i) – father*
>
> *"She is his daughter too! I don't agree with him opting out but he says I take over – I don't think I do. He lacks confidence with what she needs so I just get on with it". Sitting back she thought for a minute and then said, "I don't know if he is a bit afraid of her and it is a bit of loneliness."*
>
> *Leonora i) – mother*

Overall the impact of the child's life limiting illness changed the lives of parents in complex ways, on a daily basis and over the child's life span.

> *"The worry and the problems never go away! Never! It (the child's life limiting illness) affects all of your life ... every part of in your life ... and everything in your life." Daphne i) - father.*
>
> *"It's just the weirdest life – it is a weird kind of life!" Reuben b) – mother*
>
> *"You can't understand this life unless you live it." Reuben a) – father*

7.4 Disruption to the life stories of siblings

In all participating families with more then one child, the child's life limiting illness impacted on the life story of siblings [*Reuben a), Reuben b), Imogen c), Jagger e), Jagger g), Daphne h), Marcus j) and Marcus k)]*. Day-to-day outings and plans for siblings had to be changed if the child was ill. Young children expressed disappointment and resentment about these lost opportunities.

> *'When Daphne is in hospital I don't get to play. Sometimes I don't get to do things cause Daphne won't do it! … We were going to the circus but Daphne said 'no' so I didn't go … but mum says we might go. Alice comes on Saturday just to take Daphne out, not me!"*
>
> *Toni - Do you feel left out?*
>
> *"Yes … They go out or something … it's for Daphne! Not me …"*
>
> *Toni - Is it hard for you?*
>
> *"Yeah cause if Daphne is in hospital I can't play and stuff. If she is ill … see."*
>
> *Daphne h) – sister (aged six)*

Some older siblings reflected back on feelings of antipathy towards their brother or sister when they were children and touched on the impact on their own life of the child's life limiting illness.

> *'No well I was an only child and not only did I have a sister I had a sister with special needs. I do remember resenting her and not being nice to her. It was like two hits in one. I was no longer an only child and my sister was special! Of course you realise how precious they are as you get older … no I really love her now and in a way I did then but it did change my life … yes it did."*
> *Daphne h) – sister (aged 22)*

Parents worried about the care roles taken on by older siblings and whether the extra responsibilities changed siblings as people, as well as altering their life story.

"We really, really regret what we've done in that our daughter is a very, very lonely child. She's not a child any more she is a young woman, she has no friends of her own, absolutely none at all, she doesn't interact with anybody of her own age. We blame ourselves for it because you know it's only been recently I've been in a job where I've got reasonable money. Up until then I worked every weekend to keep money coming into the house and my wife had to rely on our daughter. Everything she had to do our daughter had to be involved and it's, you know, at the time we just sort of said, 'Oh it can't do any harm,' But actually I think it has done harm, it's taken away a lot of her childhood. I'm not sure how my daughter would have been … because she is a quiet girl anyway …but I regret her helping with caring for Jagger and maybe it has changed her. Certainly she would have had a different life."
Jagger g) – father

Although some siblings were directly involved in helping to care for the child, the extent to which they took on these roles varied from family to family and, regardless of care roles, the child's life limiting illness did have an impact on the lives of all the participating siblings. The impact varied from day-to-day disappointments to long-term changes in the life stories of siblings.

7.5 Summary

In all cases the child's identity was bound to the child's body, social roles, interactions and their life story.

Where the life limiting illness emerged early in the child's life (whether it was diagnosed or not), there was a continuity to the child's presentation of themselves that was supported and nurtured by parents and siblings. Children, parents and siblings down played any physical signs of the life limiting illness or presented them as an accepted part of the child. Although the illness, therapies, treatments and critical episodes or deterioration intruded on the child's day-to-day roles – these were presented by children and families as part of the child's daily life, not something unusual, temporary or passing – but common occurrences throughout the child's life. These children's physical appearance, social roles and life stories were integral with the life limiting condition. They were always a life limited child.

Where the life limiting illness emerged later in childhood, the child did suffer disruption to their life story: they lost their past self. Although only one of the young participants could explore this directly, parents and siblings of other children described the child who they had lost and frequently used photographs to demonstrate changes in the child's physical appearance. Although references were made to the 'lost' child, hardly any references were made to lost opportunities for the child's life or what the child's life might have been like. Instead families focused on life with the child from moment to moment and the child's personal qualities. The children and their families noted changes in the child's physical appearance and abilities but changes to the child's social roles and life story were assimilated by all participants into a new self and identity. In the case of participating families there was barely any mention of lost social roles or changes to the child's life story – rather the child's life story unfolded with the life limiting illness as part of their present and future. Disruption to the life stories of parents and siblings was however, evident and tangible in every case.

Chapter Eight

8 Synthesis and conclusions

8.1 Introduction

This is a society in which children are sometimes perceived in terms of their potential to be 'future adults' rather than 'present persons' (James and Prout, 2001). We expect children to live into adulthood and view childhood as a precursor to adulthood, personhood and identity (Hockey and James, 1993). However, not all persons make the transition to the 'age of majority' and the assumption of personhood (Hockey and James, 1993). The literature review revealed that when children suffer from life limiting conditions we fail to ask them about their thoughts and experiences. Do we undervalue the views of children who may not reach adulthood? Bluebond-Langer (1980) found that studies of socialization had failed to explore the active roles of children and focused on what children would become: 'children and childhood' only in terms of potential for the future. Her question was,

> *"How can one use a model that presumes a future, that presupposes adulthood, for children who will never reach adulthood, particularly when everyone associated with these children is acutely and painfully aware of the fact?" (Bluebond-Langner 1980 p6)*

Fraser and Robinson (2006) suggested that empirical research with children might help to uncover how social constructs like 'children', 'childhood' and 'disability' operate to maintain social power and place, not just to reveal the relative powerless place of children but also to explore the active roles of children in society:

> *"The focus of empirical research is then about how children and young people achieve 'agency'; that is, agency refers to the ability of a social actor to engage in the process of construction of meaning or identity in such a way as to influence the form that that meaning or identity takes." (Fraser and Robinson, 2006 p76)*

The purpose of this study was to hear from children affected by life limiting conditions and their families and to understand more about the following questions:

1. How do families face the dilemma of whether or how to be open about the life limiting nature of the child's condition within the family?
2. What is the impact of having a life limiting condition on a child's developing sense of self?
3. Do children who are unable to communicate verbally have active and/or passive roles in shaping their identities?
4. What are the current roles of support services like children's hospices? What are the challenges and opportunities for developing support that matches the needs of life limited children and their families more closely?

The theoretical approach to the research was bound to interpretism and constructionism and the aim of the design was to create a structure for working together, so that I would come to understand participants' views about their personal experiences through naturalistic methodological procedures in 'real life settings'. Bronfenbrenner's ecological systems theory influenced the study design and the desire to capture information about interactions and relationships between the child and others in a range of social settings. Some of this information was gathered through observation and some through the accounts of participants - for example, stories of time at school or visits to hospital.

I hoped that the study design would encourage the children especially to reveal and represent themselves in their own ways. Although the study approach was drawn towards social constructionism, questions emerged from the inclusion of children who communicated non-verbally. Given that social constructionism links the formation of social concepts to language, did the children who were unable to communicate verbally have active and/or passive roles in shaping their identities? The methods chapter contains extended reflection about the process of involving children with a range of needs and abilities in research. Overall I would say that details of the design could probably be refined but the children and siblings participating in this study responded positively to the idea of 'working' with me and having choices about how to represent themselves and their lives.

This chapter draws together and debates main themes from the findings of the study. These are compared and contrasted with seminal theories from the literature. Implications for the care of life limited children and their families are explored. Using a social construction approach I have focused on how social concepts of 'the child' and 'the life limiting illness', were constructed and presented by the children and their families. The impact of the children's and families' experiences and the wider social context of the lives of participants are also explored.

8.2 Case presentations and 'moments of realisation'

Before this study the experience of life limiting illness from the point of view of affected children and their families had not been researched and was not well understood. The conceptual cases presented here are an original and creative way to portray something to the reader about the uniqueness and commonalities I found amongst the participants and their stories. Inspired by the writings of Robert Stake (1995) I have drawn out the five stories presented in chapter four – portraits to help the reader know more about the children and their families without meeting them in person. The conceptual cases offer a level of generalisation because each one is based on evidence from more than one case. Since the findings all arise from multiple case analysis, it is hoped that life limited children, their families and those who care for them may identify with some of the themes and issues revealed by this research and understand more about their day-to-day experiences..

Little is known about the pattern of living with a life limiting childhood illness partly because the range of illnesses is wide, the consequences are assumed to be diverse and there is a paucity of research. The study identified the following 'moments of realisation' of the participating children's unfolding life stories, which represented times when families recognised the real threat to the child's life from the illness.

- Questions of inheritance, diagnosis and prognosis
- Acute loss of abilities
- Slow deterioration
- Life threatening surgery
- The cycle of crisis and survival
- The child's life and death

These moments ebbed and flowed throughout the child's life and each recurring crisis brought with it pain and uncertainty for the child and all members of their family. Sometimes there were long periods of stability in the child's state of health and at others crises came quickly or repeatedly. The child's condition could remain unchanged, be altered in the short term or there could be permanent deterioration. 'Moments of realisation' stopped life for the children and families and intruded on reality, triggering insights into immediate and future losses. Uncertainty around these times had many dimensions for families, they did not know when the next change in the child's state of health would emerge, what form it would take or the likely immediate and long term consequences for the child and family. 'Moments of realisation ' brought the need for readjustment for children and families generally followed by periods of transition and relative stability but for an unknown period of time. Uncertainty was largely unacknowledged or discussed within families.

'Moments of realisation' centred around recurring times of crisis or change in the child's state of health because these were moments of increased anxiety and fear; times when the threat to the child's life became clear to family members. This finding is supported by research, which indicated that times of deterioration in the child's health (where the child has a life limiting condition) increased anxiety and uncertainty amongst family members (Cohen, 1995; and Steele, 2000). This study suggests further that 'moments of realisation' created recurring times in the child's life story, when there was increased pressure on family members to talk to each other about the life limiting nature of the child's illness. Complexity arose because families were under increased emotional pressure at these times. They had to focus on the immediate needs of the child, were uncertain whether to talk to the child about death and dying at these moments and could never be certain whether the child would survive and live for months or years afterwards.

Glaser and Strauss (1968) highlighted how uncertainty and time can combine to create four different dying trajectories:

1. Certain death at a known time
2. Certain death at an unknown time

3. Uncertain death but a known time when the question would be resolved
4. Uncertain death and unknown time when the question would be resolved

Glaser and Strauss (1968) suggested that expectations of dying determined how everyone interacted with the dying person and each other. Expectations arose from physical clues and temporal clues about - for example, the typical progress of the disease. It was found that when the pattern of dying did not follow expectations it required a change in response that could cause havoc with systems and the 'order of work'. It was also found that unexpected changes in a persons dying trajectory altered the patterning of mood and sentiment that characteristically existed within health care settings – for example, hospital wards. Steele (2002) used the concept of 'certain death at an unknown time', drawn from the work of Glaser and Strauss, to examine the experiences of families caring for children with neurodegenerative illness and found that at times of crisis in the child's health, families felt they were 'falling off the plateau' (times of relative stability that emerged between times of crisis).

The children taking part in this study suffered from a range of medical conditions, often rare or undiagnosed – in most cases there was little information available to families or professionals about the typical progress of the disease. Families participating in this study faced prolonged and fluctuating uncertainty about the child's life and death over years. Within families, levels of certainty and uncertainty about the future course of the child's life limiting illness rose and fell amongst individual family members. Individuals introduced layers of certainty and uncertainty into their interpretation of the child's unfolding life story. Uncertainty was a recurring theme that rose and fell within the family unit and across individual family members. Themes within families followed the same pattern of 'moments of realisation' but individuals drew their own conclusions from their personal perspectives of these times in the course of the child's illness and unfolding life story.

This study suggests that there are recurring moments in the lives of life limited children when families face extreme emotional pain and are under increased pressure to talk about the child's life limiting illness with the child and other family members; times when it is difficult to make decisions about communication. The research has revealed 'times' in the child's life when families need extra support with family communication and new ways to talk to the child about their unfolding life story and life limiting illness. These times were generally followed by periods of steadiness and transition. Children and families needed practical support at times of crisis but alternative communication strategies could be introduced between these times to facilitate transition into and during periods of limited change in the child's general state of health.

Concepts of dying trajectories arose from work with adults, not children and not whole families. Whilst they have influenced thinking about dying they are somewhat linear and do not reveal the complexity for children and families of living with a life limiting illness over long periods of time, even years, combined with responding to the needs of a developing child. It is hoped that the conceptual cases and the concept of 'moments of realisation' might give professionals a new sense of day-to-day life for life limited children and their families, altering how they view potential opportunities for working with these children and their families. Figure 9 provides an overview of the study findings and illustrates how much pressure life limited children and their families face on a day-to-day basis.

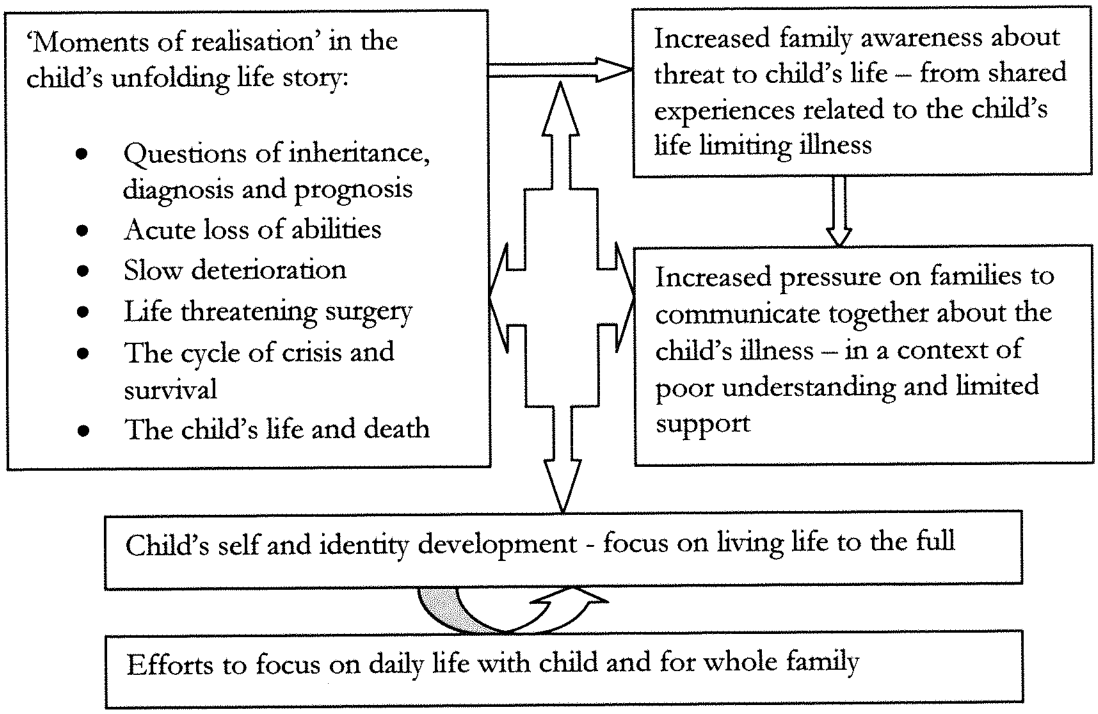


Figure 9: Pressure on family communication from the child's life limiting illness and efforts to maintain daily family life with and for the child and whole family

8.3 Family awareness and communication

Debate in the literature about communication with dying children is largely based on the knowledge of people who have worked closely with dying children but provides little or no research based evidence for professionals and parents about how to talk to dying children about death. It is suggested - for example, that there are important differences between children who are terminally ill and children who are dying (Tadmor et al, 2003). Tadmor et al (2003) pointed out that in their experience children focus on life to the last weeks or days and are only ready to talk about death close to the end of their lives. Others highlight how as a society we discuss the process of birth with children but rarely the process of dying (Adams and Deveau, 1995) and illustrate again that children with direct experiences will have questions, feel loss and deserve honesty and kindness (Adams and Deveau, 1995).

Largely influenced by the work of Glaser and Strauss (1965) there is an underlying assumption in many health care settings that it is desirable to talk about dying with dying people. In a study providing information about family communication when a child has cancer, van Veldhuizen and Last (1991) suggested that children with cancer should be informed as soon after diagnosis as possible about the disease and possible implications. This would be complex for children diagnosed near birth and impossible for children with no medical diagnosis whose general deterioration indicates they may have a life limiting condition.

The participants of this study were children who were going to lead short lives. Families shut away this fact or put it aside in order to get on with daily life and live life to the full for and with the child. The idea of awareness contexts proposed by Glaser and Strauss (1965) has much to offer in terms of understanding the complex relationships between the separate notions of awareness and communication about dying. Very young children may be unaware of the life limiting nature of their illness (closed context awareness) but this study suggests that even their levels of awareness are challenged everyday by their personal experiences. Suspicion awareness may have existed within families but the children I met tended to keep their questions about life and death for 'others' and any efforts to find out more were very subtle and measured. Although mutual pretence was evident between parents and children but as suggested by Glaser and Strauss (1965) the stresses and pressure of maintaining this approach were also tangible. Open awareness did not exist between the children and their parents and no one talked about the child's dying with them at the time that they took part in the study. It should be noted that no one knew how long these children had to live, therefore, children and families focused on life and living. In addition Glaser and Strauss (1965) found that open awareness contexts did not necessarily eliminate complexity of interaction or communication between dying people and those who were caring for them.

However, awareness contexts may be an unproductive and over simplistic way to view communication in families of life limited children. The children were living with the illness over years, rather than months or weeks. They were developing children who faced deterioration in their own abilities or state of health – a different group to the dying people studied by Glaser and Strauss (1965), a group with different needs. Parents participating in this study seemed in everyway to be doing their best to care for children that they loved. Parents who wanted to talk more openly with the child about the life limiting nature of the illness did not get professional support or advise about how to communicate differently over time, in line with the changing needs of the child and their siblings. Parents found support with communication questions difficult to find and that which existed tended to involve a professional talking with the child themselves, rather then offering new communication strategies for families to use together.

This study suggests that children begin to understand that they will die early, very gradually, and that they integrate this information into their own life story. This implies a potential move away from the notion of a single moment to raise the subject of the child's dying for professionals and family members. However, the risks of this approach might be that the child's dying is never discussed. The children who participated in this study had few, if any, opportunities to explore their losses. Loss of abilities was sometimes seen as linked to the child's life limiting illness, although loss of abilities and dying are separate entities the children were offered limited chances for exploring their own reactions to either.

Although the insight of individual children should never be assumed, beginning with questions about how much the life limited child is aware of could provide a more helpful starting point for responding to needs. If we are open to hearing children's questions, we could move on to look at how to facilitate communication about children's illnesses and dying (when the times comes) especially within the family. Vygotsky's ideas about the role of society in providing a framework in which the child's active construction of meaning develops with the support of others might be very helpful here.

The efforts of participants to talk about the child's life limiting illness especially at critical times generated the following questions:

- How can parents inform a child or other family members, when information about the child's illness and prognosis are limited or ambiguous?
- How can families evolve the story of the child's illness for a developing child/young person and their siblings?
- What is the right time for families to talk about the life limiting nature of the child's illness particularly with the child?
- When is a child 'dying' and would that moment be too late for parents to talk openly with the child about their own death?
- How can professionals best support the people most likely to talk to a child affected by a life limiting illness (parents and carers or other close family members like siblings)?

The complexities for families who decided not to talk 'openly' within the family about the life limiting nature of the child's illness raised these points that should be considered in efforts to support these children and their families:

- The child's life limiting illness is not one subject; experiences necessitate talk between the child and parents or siblings at different levels and at various times - for example, about treatment procedures;
- Physical evidence of the illness on the child's person can play a role in revealing the 'truth' to the child and their siblings; this might include scars, changes in the child's body or altered abilities;

- Invariably experiences related to the child's illness or treatment are shared and can raise levels of awareness about the threat to the child's life within the family, even if events are not discussed;
- Children and siblings make independent evaluations of what they experience and hear outside the family home and parents wishing to protect the child do not so easily control levels of awareness in other social settings.

Overwhelmingly the children, parents and siblings lived with the pain of shared experiences related to the child's life limiting illness: frequent hospital trips, sudden and gradual changes in the child's health status. The whole family struggled with questions about how to communicate within the family about the child's illness, whilst trying to maintain stability for the child and all family members through daily family life. The problem for families was how to deal with all the complex and varied pressures of family life with the child's life limiting condition as a constant added threat and force. A presence they were all painfully aware of (often through shared experiences rather than talk) but unsure how to communicate about. Decisions related to communication within families were largely influenced by concerns held by the child and/or family members about how to communicate without harming each other. There was interplay between families' efforts to manage talk about the life limiting nature of the child's illness and the intrusion of personal experiences. This was especially the case in settings outside the family home, where the child was away from parents and drew their own conclusions about differences between themselves and school peers, interpreted interactions and experiences in health and social care settings and wondered about social meanings of the word hospice.

Participants talked about the child's life limiting illness by separating it into three parts:

i) The illness – all family members talked in general terms to each other about the child's illness, symptoms and therapies without reference to the threat to the child's life;

ii) Death in childhood – only the children affected by life limiting conditions raised the subject of death in childhood through the research process, they used literature and media stories to bring up the general topic of children dying;

iii) The child's dying – in participating families parents often talked to siblings about the life limiting nature of the child's illness; I did not meet any families where this was discussed with the child; one young person used metaphor to raise the subject of her own dying but she was met with hesitation because no-one wanted to say more than she was ready to hear.

Parents who were unable to talk about the prospect of their own child's dying were able to explore the deaths of 'other' children that they witnessed in health care settings. Figure 9 examines the idea that study participants have highlighted a number of potential 'bridging topics' that might facilitate talk within families about the child's life limiting illness.

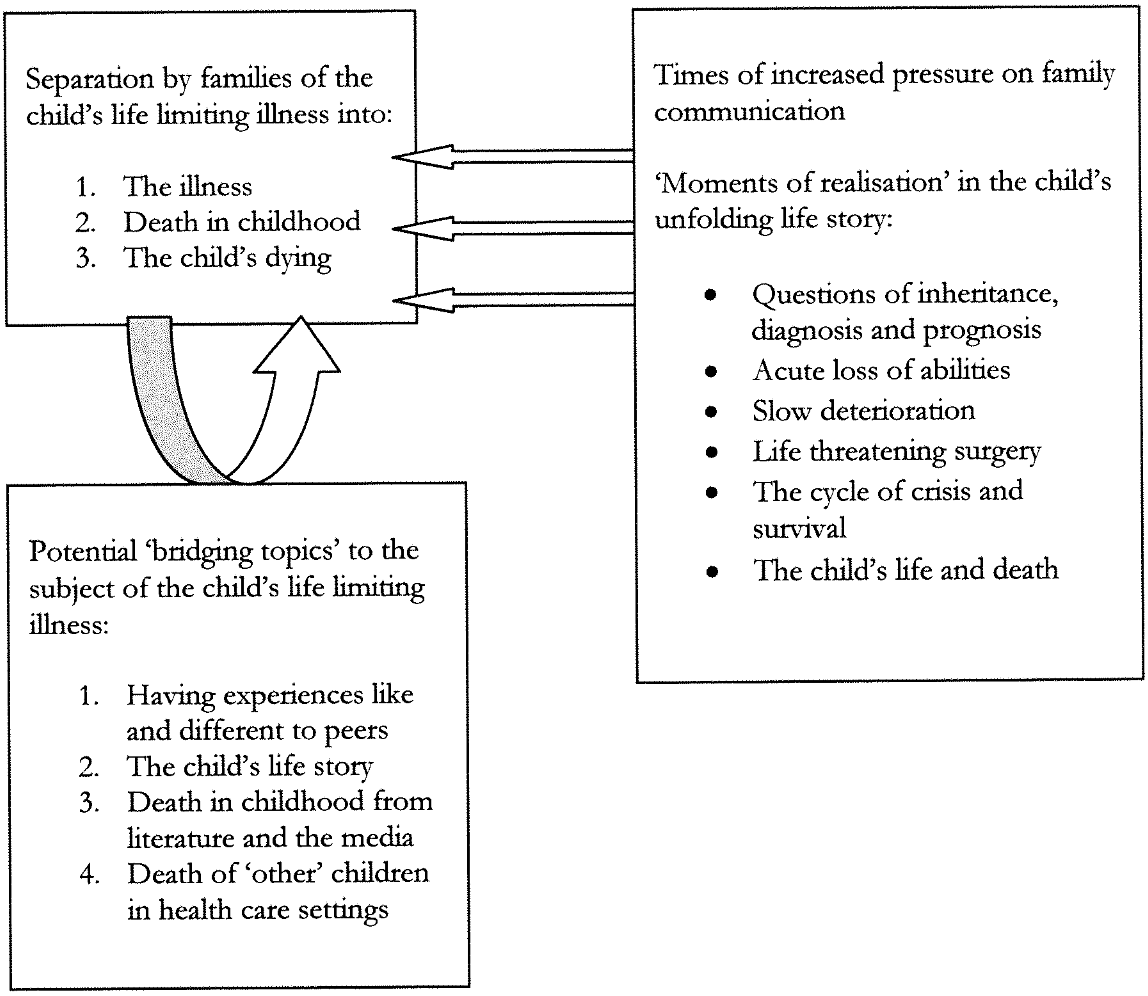


Figure 10: Recurring pressure on family communication and potential bridging topics to facilitate talk about the child's life limiting illness

Participants divided the subject of the child's life limiting illness into three parts through participation in the research and potential 'bridging topics' emerged from analysis of the subjects used by participants to raise subject like 'death in childhood' or the 'deaths of other children' with me (the researcher). These topics might help families of life limited children to talk together about the child's life limiting illness by division of a painful subject into smaller and less direct topics.

8.4 Self, identity and biographical disruption

The 'self' is said to be a multifaceted and complex social concept. The self is a private theory and therefore difficult to interpret from outside. Understanding the self-concept of another person depends on the extent to which we share ourselves with others. Schaffer (1996) suggested that the self is a 'theory' people develop during childhood about who they are and how they fit into the society within which they live. Central to how we each construct reality is our understanding of ourselves. It is argued that the theory of self is not static but builds up throughout life and is most vulnerable to experience in childhood when we continuously compare ourselves to our own self image, to others and to idealised images and how we perceive other people's reactions to us.

> *"The function of the theory (of self) are to help us seek out particular experiences that fit in with our self image, make choices among alternatives courses of action, and experience pleasure or displeasure in evaluating the results of such actions. The theory is a complex one; it contains a great many constituents (self-awareness, self-concept, self-esteem and so forth) all of which need to be knitted together, and it is characterised by both cognitive and emotional features." (Schaffer, 1996 p169)*

Schaffer (1996) described how very young children begin with self-recognition through visual images in mirrors or video, use of the words 'I' or 'me' and recognition and response to their own name. Young children often have a basic 'either' 'or' self-concept and only at age seven or eight do children compare themselves to others, beginning with appearance.

The thesis focused on four theories of child development Erikson in particular highlighted personality and identity development particularly during adolescence and his theory confines reflections on life and acceptance of the inevitability of death to adulthood. Piaget hypothesised that standard cognitive development occurred in stages bound to chronological age. Later research went on to utilise Piaget's stages of cognitive development to hypothesise that children classically come to understand death and dying in stages tied to chronological age. Although research into the perceptions and experiences of dying children is very limited there is evidence to suggest that personal experiences can lead children to have a more developed understanding of dying than might be anticipated for the average child of their age (Adams and Deveau, 1995; Bluebond-Langner, 1980; Davies, 2002; Doka, 1995; Hockley, 2000; Judd, 1995; Matthews, 1994).

The literature review suggested that the physical needs of 'ill' children can upstage the child's developing self by highlighting differences between the child and peers (Samaniego et al,1977; Wise, 2002; Woodgate and Degner, 2003; Mullis, Mullis and Kerchoff, 1992; Walker et al, 1999; and Snethen et al, 2001) and that 'the body' should be considered in theories of dying (Copp, 1999).

The role of the child's body especially in disability is rarely considered in theories of child development; it is taken for granted. The disabled child's body, especially where they have a visible disability which makes them look different, affects how they are perceived and responded to by others and is inevitably part of who they are as a person. The life limited children taking part in the research lived every day with a range of physical differences between themselves and their peers and they appeared to subsume physical differences into a rounded presentation of self in which physical differences were not placed centre stage. The children actively able to represent themselves and the families of children unable to represent themselves wanted me and others to see the person beyond the needs of the child's body, beyond the physical presence of the child. However, the children taking part in this research lived not just with disability but with deterioration and physical changes in the their body and abilities that played a role in revealing the 'truth' about the life limiting nature of the child's illness.

The following factors influenced the participant's efforts to protect and present the children as a unique individuals, each factor had a role in revealing the truth about the child's vulnerability that was beyond the control of families through talking and not talking efforts:

1. The child's body, growth and development, visible and invisible disability, scars from surgery or treatments and other signs of the child's illness - for example, jaundiced skin
2. The child's social roles, interactions and experiences especially away from family and outside the family home
3. The child as an active agent seeking and interpreting information and experiences independently
4. The role of family in protecting and presenting children who were unable to represent themselves

All children have to gain an understanding of death and dying as they grow and develop. Researchers have used the work of Piaget to develop classical theories about how children develop concepts of death in stages. People studying or working with children who have direct experiences of death and dying, increasingly challenge the idea of stages tied to chronological age. The children studied by Bluebond-Langner experienced accelerated acquisition of knowledge about their own dying due to the process of their illness, treatments, treatment settings and observation of children dying of the same illness. The children with leukemia studied by Myra Bluebond-Langner (1980) moved from a self-concept of being a well child through other self-concepts to being a dying child. The children's concepts of self changed with information and experiences, the children's self-concepts were especially influenced by what they observed about other children with the same diagnosis who they met and interacted with over the course of their illness at the treatment centre. It appeared from Bluebond-Langner's study (1980) that the children with leukemia who participated were watching other children knowing there would be implications (things to learn) about themselves and the likely course of their illness.

The children taking part in this study rarely met a child with the same diagnosis, the hospice was an infrequent place that they visited (about 15 nights per year) for fun and rest rather than any active forms of treatment. I did not find that they watched each other and largely they only mentioned other children at the hospice in terms of social interactions. The self-concepts of the children taking part in this study seemed to alter very gradually through awareness of changes in their own body or physical abilities and by comparison of their own experiences and social roles with those of their peers. Information about the life limiting nature of their illness came to them gradually over the span of their lifetime and was integrated by the children into the story of their own lives.

Bluebond-Langner (1980) found that deterioration in the child's health came through a series of relapses. Early relapses brought changes in treatment but all relapses were revealed to the children in sudden changes in the reactions of others. The children became increasingly aware that the adults around them began to offer less information, interaction and honesty. Although some of the children taking part in this research suffered acute phases of their illness and sudden deterioration, slow deterioration was more common and it emerged almost invisibly over long periods of time with barely any discernable changes in treatments, therapies or the reactions of others. The child's identity and developing sense of self was inextricably bound up with their life limiting illness. The child's life story was framed and shaped by the expectation of family and others involved with their care that they would lead a short life. Their bodies revealed signs of the illness and the illness intruded on their social interactions. The children taking part in this study used this range of information to monitor, evaluate and form concepts of themselves and their worlds. Many aspects of the child's sense of self seemed to indicate to them that they were different to other children of the same age but most challenging to the child's identity (as explained by their family) and sense of self (as explained by the child) have been described as 'moments of realisation' in the child's unfolding life story, because these moments revealed the threat to the child's life.

Chronic life threatening illness in adult years has been said to lead to 'biographical disruption' (Bury, 1982) in that it changes people's life course and personal identity both in their own eyes and in the eyes of others. Chronic illness was said to disrupt everyday life, social relationships and the forms of knowledge underpinning modern life. The biographical disruption seen as a consequence of chronic life threatening illness in adulthood seemed to apply to participating parents and siblings. This finding supports the work of Young et al (2002) who also found that the life stories of mothers (in that study) were utterly disrupted by their child's illness so that assumptions about daily life and life in general were brought into question and could no longer be taken for granted.

Others have found alternative or parallel concepts of 'biographical continuity' and 'biographical reinforcement (Levealahti, Tishelman and Ohlen, 2006; Carricaburu and Pierret, 1995). Williams (2000) reviewed concepts of biographical disruption and argued that studies had so far failed to take account of situation in which the illness could be a central part of biography from birth.

This study highlighted the remarkable capacity of children to integrate a life limiting illness into their life story and live life to the full. Children whose life limiting illness is diagnosed early in babyhood have an identity within which the life limiting illness was integrated throughout their lives. The illness and consequences seemed to be integrated into the child's sense of self and identity. The children I met did not 'redefine' themselves because of their illness. For them, self-definition work had to incorporate the life limiting illness because they were growing children, some of them very young, all of them hardly able to remember a time before their illness.

Where the child's life limiting illness emerged later in their childhood, participants did describe loss of the child before the illness. I only met one young person who was able to articulate memories of who she was becoming in the time before her illness. Parents and siblings articulated these memories for children less able to represent themselves. For this participant I would say that the concepts of biographical disruption and redefinition of self did arise from her illness to some degree. However, even her memories were confined to one aspect of her life and physical skills, not overwhelming who she had become since the illness and assimilated into her new identity. This finding tends to support the ideas of Levealahti et al (2006) related to 'biographical continuity' and 'biographical disruption' on a continuum where – for example, gradual bodily changes can be integrated into the person's biography.

The families lived day-to-day in a society that expects children to live into adulthood and where disability is looked at in judgemental and superficial ways but is not discussed or fully understood. The families taking part in this research largely felt alone with the knowledge that their child would die young. Although elements of social support were praised and individual professionals were valued, overall practical and emotional support seemed to be inconsistent. Families were aware of the wider social context within which they lived indicated by: talk of Government league tables and political pressure on health and social care providers to spend money on 'viable' children. Particular talk about the lack of social resources for support for the child and whole family in their own home or locally and the emphasis on charitable agencies rather then services with any statutory funding were all common themes. Families talked about social 'staring' in terms of feeling judged. Families expressed the notion that judgements were made about the child as a valuable person, their right to life and their family. Children and siblings especially found these kinds of judgements painful and difficult to cope with and reacted with resentment and anger. Participants felt that disability was still a social taboo and the child's life limiting illness was not open for debate in social groups outside of their immediate family.

8.5 Conclusion and implications for care

The conceptual cases have revealed a series of 'moments of realisation' in the life stories of life limited children: times when the life limiting nature of the child's illness is most clear to families, which heightened uncertainty and the need to talk.

The concept of potential 'bridging topics' to facilitate talk in families caring for children affected by life limiting conditions has two implications for the care of life limited children and their families:

1. Professionals should be aware that 'moments of realisation' in the child's unfolding life story might highlight times in the child's life when pressure on family communication is increased;
2. Children and families may find that the 'bridging topics' utilised by participants of this research could facilitate talk within the family about the child's life limiting illness through indirect paths.

It might help families of children affected by life limiting conditions if professionals could explore with them how to make talk and awareness more manageable perhaps by breaking it into smaller components. Overall it might be helpful to be open to talk within the family about the child's whole life story, including a wide range of personal and family memories, of good times as well as events related to the child's illness. For example:

- Discuss the child's life story in the form of anecdotes and family memories

- Celebrate the child's sense of self and identity acknowledging the integrated signs of the illness on their body, social roles and life story
- Celebrate the child's past and present identity acknowledging the changes brought about by the onset of the illness on their body, social roles and life story
- Talk about the child's diagnosis in general and non-specific terms
- Talk about the child's or shared family experiences in specific health/social care settings on a day-to-day basis
- Acknowledge that certain experiences are shared but painful or difficult to talk about
- Acknowledge that awareness and talk might not match or be in balance
- Try to talk about 'moments of realisation' in the child's life story, gently exploring emotions within the family without making the child feel responsible for the reactions of other family members but acknowledging that these times are painful and difficult for all family members
- Talk about death and dying in general
- Talk about death in childhood
- Talk about the child's life limiting illness
- Talk about the child's dying, if the moment arises and those involved feel able to respond honestly to the child's questions

Exposing the complexity of the child's life limiting condition may help children, families and professionals to take steps towards talking about the experiences, emotions and day-to-day consequences of the child's life limiting illness for the child and all members of their family. Professionals might be able to use this idea of complexity to create manageable aspects of talk to facilitate talk between children and their families. Eventually this could cascade into other environments and influence awareness about life limiting illness and death in childhood, through links and processes across social settings.

The study also suggests that in terms of self-concepts and identity formation, the children taking part integrated their life limiting illness into concepts of themselves and accepted their short biography. The children lived every day with the consequences of their illness and it seemed as if information about the life limiting nature of their illness came to them very gradually over the course of their life span. This was also true for children whose illness emerged later in childhood; although they and their families had memories of the child before the illness, changes were assimilated into a new identity. The children challenged concepts of biographical disruption because they presented such continuity in their representation of themselves. Parents and siblings however highlighted the overwhelming impact of the child's illness on their own lives and their personal biographical disruption was evident.

8.6 Study limitations

Like all research, the study was limited by certain aspects of the design and methods leading to the potential for bias. Care team members from the children's hospice identified and recruited families to take part in the research, because this seemed the best way to ensure that the selection criteria were considered and to protect families facing any immediate problems that I would not have been aware of. However I have no certainty about the true rationale or motivation of staff for choosing particular families to approach about the study over any other families supported by the hospice service. There is a possibility that care team members consciously or unconsciously chose families that they found easiest to approach or that might have seemed most positive about the service. However I was unable to access or recruit families to the study any other way.

It is difficult to assess the extent to which families might have felt obliged to take part in the study, given that the approach was made by staff from a service that is provided free of charge and for vulnerable children, and families who in some cases received limited support from any other agencies, especially in terms of respite breaks. I made every effort to ensure that families were reassured that any decisions about participation or withdrawal from the research would have no impact on the care of their child or children. I was somewhat reassured that three families (one for the pilot study and two for the main study) either declined to participate or failed to 'get back to' the care team member who approached them about the study. I was also reassured that three teenage brothers and two fathers from separate families either declined to take part or were always unavailable when I visited the family homes. Although no child or any of the younger siblings declined to take part, the methods included a range of strategies to make the process of consent an on-going one. To enable the children and their siblings to have some control over the time, length and location of sessions with me, I practised with them how they would let me know if they had enough and wanted to stop each session at any stage.

I was aware of the possibility that children, siblings and parents might feel obliged or inclined to 'say the right thing' especially about the hospice service. Besides inviting participants to represent themselves in their own ways, I was asking them about their wider day-to-day experiences, not just about support from the hospice. I found that parents particularly would talk at length in positive terms about the children's hospice and I would listen, acknowledge how positive they were about the support provided and then ask, "Was there any aspect of support that did not work for you, your family or your child?" I did find that most participants could and did identify aspects of support from a range of agencies that did not meet the needs of their child and family in some way.

It is not clear to what extent the participants might represent other life limited children and their families. The general limitations of case study research have been discussed in the methods section but it as hoped, as Stake (1995) suggested, that readers have glimpsed into worlds they would not otherwise have seen.

At the outset of this study I worried about whether I had the right personal qualities, skills or abilities to involve children in research. As I said at the start of this thesis it was good advice to myself to make no judgements about work with the first child to participate in the study until we had met each other two or three times – I reiterate that this was good advice to myself and to others who want to involve children in research but who are not sure if 'they are the right person for the task'. I believe that if you offer children an honest explanation, time, and a sincere effort to listen to their thoughts and experiences, they will make their own decisions about who you are and how much to share with you; but they will share with you.

My role with the children and families was a fleeting one, intense for us all during the study but one we knew would be short. I always had the feeling that the children and families were as certain of this as I was; like lots of other professionals I was a passing presence, someone who would move on. The families decided to take part in the study for different reasons which we did not necessarily discuss but many said they participated to increase awareness about the experiences of life limited children and their families, over long periods of time and on a daily basis, they have entrusted me with this task. I see some of the children and families when they visit the hospice and we stop to pass the time of day but most never mention the research. I am not certain that any of the children and families will want details of the findings, when I offer them – I believe they have moved on with their lives and are leaving the task of raising awareness to me.

It is almost impossible at this point in time to separate the impact of the research on me from my own losses and bereavement. At the end of the day our own losses cut more deeply than the losses of others, no matter how much we empathise. I had and have the support I needed to undertake this study. I am reminded of one of the siblings who was telling me that other people should not listen to the story of her brothers life and cry – they should listen and hear what life is like for their family every day and think about what would help,

> *'They don't want to sit and cry about it all - they want to listen and think about how to help us, what we need." Jagger e) – sister (aged 14).*

I truly hope that this thesis begins to make you do that.

8.7 Further research

The study raises interesting questions about how the experiences of people affected by life limiting illness over months or years might be better understood. There might be opportunities to develop concepts beside the idea of 'illness trajectory' to facilitate a better understanding of the impact of illnesses that will lead to death and shortened life expectancy; but over long periods of time and life with all the consequences of the illness for the affected individual and their family.

The study has brought forward questions about how participating families tried to manage to talk about the life limiting nature of the child's illness, especially with children who were cognitively able and who communicated verbally. However, further work could be undertaken to find ways of helping families to 'talk' and respond to the changing needs of the developing child but also finding ways to take into account the role of the child's personal, particular experiences, especially those related to or influenced by their illness.

The implications of the research in the broadest sense indicate that socially there is a poor level of understanding about the complex needs of children affected by life limiting conditions and the consequences for their whole family. There is scope for further research into the needs and experiences of life limited children and their families, perhaps using a range of qualitative and quantitative methods to produce more generalisable results about specific questions. Suggestions for future research arising from this study include:

- To explore the concepts of 'moments of realisation' and 'potential bridging topics' with professionals caring for life limited children and their families – looking at how they interpret these concepts and implications for care.

- An examination of social reactions and interpretations of the terms life limited, life threatened and hospice – in relation to children.

- An exploration of the similarities and differences between the experiences of life limiting illness in adulthood compared to life limiting illness in childhood.

- To hear more from life limited children about how they understand, live with and experience their illness.

- To develop methods for understanding the experience of life limited children who communicate non-verbally.

- To explore the experiences of siblings of life limited children in more detail

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9 Appendices

- **Table of Research Evidence**
- **Table of Discussion Papers**
- **Outline of ethics process of redesign and review**
- **Pilot Study Table: Analysis of Pilot Data**
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TABLE OF RESEARCH EVIDENCE

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Aitken, T	Tamara J Aitken and Gay Hathaway. Long distance related stressors and coping behaviors in parents of children with cancer. Journal of Pediatric Oncology Nursing 10 (1):3-12, 1993.	53 parents of children with cancer	To investigate the expereinces of parents of children with cancer.	Quantitative	Parents given 40 minutes to complete the questionnaire in a quite room. Tested questionnaire used plus data sheet created for the study.	Authors found that parents living further away, lived in rural areas, were poorer and less well educated than the group living nearer the treatment center. These differences confused the findings, i.e. all was not related just to distance from treatment.
Aylott, M	M. Aylott. Interviewing as therapy: researching parents' experiences of their child's life-threatening illness requiring ECMO. Nurs Crit Care 7 (4):163-170, 2002.	8 sets of parents	To explore the retrospective accounts of parents about their child's critical illness requiring extracoporeal membrane oxygenation (ECMO).	Grounded theory	Retrospective in depth interviews.	The parents were said to have found participation in the study therapeutic. This was described as especially important because of the parent's sense of social isolation, feeling unable to talk to friends, relatives or even each other for fear of causing up-set or pain. The author suggests that more research is needed to understand the impact of participating in qualitative research.
Barrera, M	M. Barrera, C. F. Fleming, and F. S. Khan. The role of emotional social support in the psychological adjustment of siblings of children with cancer. Child Care Health Dev. 30 (2):103-111, 2004.	47 siblings of children with cancer referred for behaviour problems. 25 siblings of children with cancer not referred for support. The siblings were aged 6-18 years.	To examine the role of emotional social support in the psychological adjustment of siblings of paediatric cancer patients.	Quantitative	Children's Depression Inventory. Anxiety Inventory for Children and behaviour. Sibling Perception Questionnaire. Parents STAIC-Parent Form. Child Behaviour Checklist and provided demographic details.	Siblings who perceived that they had access to good social support reported less emotional/behaviour problems. Parents of referred siblings reported more behaviour problems for the child. Referred adolescent females reported greater depression then referred adolescent males or non referred adolescent females.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Bertoia, J	J Bertoia. Drawings from a dying child: Insights into death from a jungian perspective, London:Routledge, 1993.	One nine year old girl dying of leukemia	To understand the 'inner experience' of a young girl dying of leukaemia, evaluated from her drawings.	A longitudinal case study	Analysis of the child's drawings. Study of medical records and interview with the child's mother including use of information in the mother's diary.	It was found that many of the child's drawings could be matched to Bluebond-Langner's stages of awareness in dying children, when the drawings were assessed in groups and alongside other supporting information. The child's parents and other carers also expressed the idea that she had pre-conscious or 'spritual' awareness of her own dying, outside the stages described by Bluebond-Langner. The author compared these notions to the ideas of Kubler Ross and Bach. It was argued that Bluebond-Langner's model based on behaviour and intellectual processes did not include the 'inner spiritual' experience of the child expressed through the 'non-verbal symbolic language' of her drawings.
Bluebond-Langner, M	M Bluebond-Langner. The private worlds of dying children. Princeton, New Jersey:Princeton Paperbacks. 1980.	32 children with leukemia, 6 acting as primary informants. The children were aged 3-9 years.	To study awareness and communication in terminally ill children.	Ethnography	Ethnography. Modified play therapy with no direct questioning.	The study explored how children come to know they are dying when no one tells them and how they conceal this information from parents and medical staff. It was found that the children were supporting others in their social roles by concealment and pretence. They discovered their prognosis whether told or not, in a complex way over time. The children also conceptualize the illness differently over time from acute to fatal and their experiences were more central to their coming to know then their ages or stages of development.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Bluebond-Langner, M	M Bluebond-Langner. In the shadow of illness: parents and siblings of the chronically ill child, New Jersey. Chichester. West Sussex.:Princeton University Press, 1996.	40 families of children with cystic fibrosis were the main focus. They were selected from a wider group of 175 families interviewed at an earlier phase of the study.	To examine the impact on parents and siblings of children suffering from cystic fibrosis	Ethnography	Field notes made by the researcher at all stages of data collection. Audio taped interviews and conversations. Observation.	The researcher argued that the impact of a child's chronic life threatening illness on parents and siblings could only be understood through exploration of 'the fabric of family life'. It was found that from the moment of diagnosis families worked to contain the 'intrusion' of the illness into their lives for as long as they could. Parents were said to use strategies of: 1. Routinization of CF treatment related tasks, 2 compartmentalization of information about the child's condition, 3. Avoidence of reminders of CF and its consequences, 4. Redefinition of normal, 5. Reassessment of priorities, and 6. Reconceptualization of the future. It was found that siblings based their reactions on evaluation of the responces of parents and the affected child. Siblings and parents used 'the natural history of the child's illness' to locate events in their own lives and retelling of life and everyday events.
Bossert, E	E. Bossert and I. M. Martinson. Kinetic Family Drawings-Revised: a method of determining the impact of cancer on the family as perceived by the child with cancer. J Pediatr Nurs 5 (3):204-213, 1990.	40 families including 8 children who drew 18 pictures	To analyze Kinetic Family Drawings-Revised created by children with cancer to understand their perceptions of the family's interactions while subjected to the stress of childhood cancer.	Five year longitudinal study	Kinetic Family Drawing-Revised. Children are asked to " draw a picture of everyone in your family, including you, doing something".	Part of a 5 year longitudinal study 'Impact of childhood cancer on the child and family'. Scores for communication, self-image and emotional tone increased over time to indicate increase in the children's negative perceptions of self and family interaction.
Cairns, N	N. U. Cairns, G. M. Clark, S. D. Smith, and S. B. Lansky. Adaptation of siblings to childhood malignancy. The Journal of Pediatrics 95 (3):484-487, 1979.	71 healthy siblings of children with cancer	To explore the impact of childhood cancer on school age siblings.	Quantitative	Piers-Harris Children's Self Concept Scale. The Bene-Anthony Family Relations Test. Thematic Apperception Test.	Siblings found to have more distress than patients regarding their social isolation, perception of parents as overindulgent/protective of the sick child and fear of confronting negative feelings within the family and fear of failure (among older siblings). Therfore it is argued that siblings like affected children faced severe stress when confronted with chronic life threatening illnesses like cancer.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Carnevale, F	F. A. Carnevale, E. Alexander, M. Davis, J. Rennick, and R. Troini. Daily living with distress and enrichment: the moral experience of families with ventilator-assisted children at home. Pediatrics 117 (1):e48-e60, 2006.	Participants included children, parents and siblings (38 family members) from 12 families of children requiring mechanical ventilation at home.	To understand more about the moral dimensions for families caring at home for a child requiring mechanical ventilation.	Qualitative	Richard Zaner's interpretive framework for analysis of interviews and home based observations.	Six principal themes were identified: 1. Confronting parental responsibility, sometime overwhelming. 2. Seeking normality through routines and trying to maintain a stable family home life. 3. Conflicting social values, believing that the child's life was devalued by society. 4. Living in isolation, with inadequate medical, social support or respite. 5. What about the voice of the children, the researcher found that the children and siblings were 'generally silent when asked to talk about their experiences'. 6. Questioning the moral order, families described their lives as unfair but felt they were enriched by the experience of knowing and caring for the child.
Carpenter, H	F. A. Carnevale. Revisiting Goffman's Stigma: the social experience of families with children requiring mechanical ventilation at home. J Child Health Care 11 (1):7-18, 2007.	Participants included children, parents and siblings (38 family members) from 12 families of children requiring mechanical ventilation at home.	The findings from a study of the moral experiences of children requiring mechanical ventilation were re-examined using Goffman's ideas about stigma.	Grounded theory	Re-analysis of data from interviews and home visit observations	Using Goffman's ideas about stigma the researcher found that parents were distressed by the ways that their child was undervalued socially. This was said to be influenced by the child's 'unusual' physical appearance. Repeated review by professionals of whether or not to sustain the child's life upset parents. The children and their families were said not to conform to standards of 'normality'. Parents were said to work to create a 'protective capsule' or a social space within which the child could feel 'normal'. The children were found to employ 'passing' strategies so they could mix with peers. Goffman used the term 'passing' to describe efforts to control information so that a person with stigmatic characteristics could pass as normal. Parents were also found to mediate with and for the child to fit in and be accepted socially.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Carricaburu, D	D Carricaburu and J Pierret. From biographical disruption to biographical reinforcement: the case of HIV - positive men. Sociology of Health and Illness 17 (1):65-88, 1995.	44 HIV positive men	To explore the daily consequences, meanings given to being HIV positive and the impact on identities.	Qualitative	In-depth interviews	It is suggested that participants reconstructed their personal and collective pasts and recomposed identities providing continuity of their biography. Identifies were not just reconstructed they were reinforced.
Charmaz, K	K Charmaz. Loss of self: a fundamental form of suffering in the chronically ill. Sociology of Health and Illness 5 (2):168-195, 1983.	57 chronically ill people with various diagnoses	To examine the loss of self experienced in chronic illness	Qualitative	Analysis of data from a previous study	It was found that loss of self was a fundamental form of suffering in chronically illness. It was found that people were leading restricted lives, experiencing isolation, being discredited, and burdening others. It is suggested that whilst chronic illness creates dependence on others for self definition, relationships become more problematic.
Cohen, M	M. H. Cohen. The stages of the prediagnostic period in chronic, life-threatening childhood illness: a process analysis. Res Nurs Health 18 (1):39-48, 1995.	21 parents of children with chronic life threatening illness	To explore family behaviour when a child is diagnosed with a chronic life threatening illness.	Grounded theory	Semi-structured interviews.	The study examined the pre-diagnostic reactions of parents to cues that their child was ill until the point of medical diagnosis. Initially they found explanations grounded in their everyday experiences and were reacting to subtle symptoms (Lay Explanatory STI), (STII Legitimating) came when the parents could no longer find normal explanations and sought medical help, (STIII Medical diagnostic) until medics reached and confirmed a diagnosis.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Cohen, M	M. H. Cohen. The technology-dependent child and the socially marginalized family: a provisional framework. Qual Health Res 9 (5):654-668, 1999.	4 families of children requiring ventilation (not before this admission to hospital) and expected to remain technology dependent for 1 year and not considered 'terminally' ill at the time of discharge.	To develop an explanatory model of the transition of technology dependent children to home and community based settings.	Grounded theory	Participant observation, interviews with families, health professionals, community informants, videotapes, relevant literature, other documents.	Family social status suggested to be a category that can account for significant variation both before and after discharge. Theoretical relationships existed between family social status and the concepts 'moral distress of professional staff during discharge planning' and 'social trajectory of families'. At times professionals had to consider discharge of technology dependent children home to families where they feared the family did not have sufficient social and emotional resources to cope. This was in part a reflection of limited discharge options. However the study also found that mastering the complex skills required to care for the child offered some 'marginalized families' enhanced self-esteem.
Copp, G	G Copp. Facing impending death: expereinces of patients and their nurses. Nursing Times Books. 1999	12 case studies of dying patients and their nurses	To gain insight into the expereinces of dying patients and their nurses		Participant observation and interviews	The study identified three major themes of patients; protecting and controlling, watching and waiting, holding on and letting go. Four models of dying were proposed; person ready - body not ready, person ready - body ready, person not ready - body ready and person not ready - body not ready. It was argued that the 'body' should always be considered in theories of death and dying.
Corden, A	A Corden, R Sainsbury, and P Sloper. When a child dies: money matters. Illness, Crisis & Loss 10 (2):125-137, 2002.	Bereaved parents whose children had support from the children's hospice	An exploratory study conducted in response to the belief of staff from a children's hospice that for many parents the death of their child had difficult financial impacts	Qualitative	Semi-structured interviews.	Argues that bereaved families face loss of benefits, funeral costs and extra difficulties at the time of the child's death.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Corden, A	A. Corden, P. Sloper, and R. Sainsbury. Financial effects for families after the death of a disabled or chronically ill child: a neglected dimension of bereavement. Child Care Health Dev. 28 (3):199-204, 2002.	16 families of children supported by a children's hospice before they died.	To investigate the financial circumstances of families whose child had died after a long-term illness and the factors contributing to financial difficulties.	Qualitative	Semi-structured interviews.	All of the bereaved parents studied suffered loss or reduction of social security benefit, immediate reduction in income as much as 72%, extra costs of paying for funerals and headstones where difficult to meet. Some parents were in debt following years as carers. Re-employment was a difficult and slow process. The study highlights the impact on families of caring for disabled children and illustrates how families continue to face financial difficulties after the child dies.
Davies, B	B. Davies, M. Gudmundsdottir, B. Worden, S. Orloff, L. Sumner, and P. Brenner. "Living in the dragon's shadow" fathers' experiences of a child's life-limiting illness. Death Stud. 28 (2):111-135, 2004.	8 bereaved fathers of children (aged 3 months to14 years) cared for by a paediatric hospice at home service in the USA. The children's diagnoses were: cancer (5), spinal muscular atrophy (2) and Tay Sachs (1).	To retrospectively explore the experiences of bereaved fathers whose children received support from a home based hospice program.	Retrospective grounded theory study	In depth interviews. Retrospective reflection.	Father's experiences are described in methaphoric terms of battling 'the dragon' being 'defeated' - uncertainty, responsibility, and everyday disruption (described as living in the 'dragon's shaddow'). It was found that hope for 'survival' was only relevant to those fathers whose children had cancer. The fathers of children who suffered from degenerative illnesses like Tay Sachs knew from the moment of diagnosis 'that their children would not survive the illness' and they did not have hope for survival. However they did have hope for alleviation of suffering and length of life for their child.
Davies, R	Ruth Davies, Bryn Davies, and Jo Sibert. Parents' stories of sensitive and insensitive care by paediatricians in the time leading up to and including diagnostic disclosure of a life-limiting condition in their child. Child: Care Health & Development 29 (1):77-82, 2002.	Families of children diagnosed with life-limiting conditions (14 couples and 16 mothers)	To explore parents experiences of care by paediatricians in the time leading up to and including diagnostic disclosure of a life limiting condition in their child	Qualitative	In depth interviews.	Parents accounts centered on their need to have their concerns heard and validated by paediatricians. The parents identified sensitive and insensitive practices of paediatricians. Paediatricians who were percieved by the parents as sensitive were said to combine 'technical skills' with understanding. Insensitive care in some cases was linked to long delays in diagnosis.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Davies, R	R. Davies. Mothers' stories of loss: their need to be with their dying child and their child's body after death. J Child Health Care 9 (4):288-300, 2005.	10 bereaved mothers	To compare the care of children and families who died at home, in hospital and in a children's hospice.	Qualitative – phenomenology	Single open interviews were carried out in the family home.	Four main themes emerged which were: 1. Time to be with their dying child. 2. Space and privacy to be with their dying child. 3. Time to be with their child's body after death. 4. Space and privacy to be with their child's body after death. Mothers maintained bonds with the child through memories of the child's life and death.
De Graves, S	S. D. De Graves and S. Aranda. Exploring documentation of end-of-life care of children with cancer. Int J Palliat Nurs 8 (9):435-443, 2002.	Records of 18 children aged 2-17 years, who died of cancer in an Australian hospital during 1999.	To explore documentation of end of life care for children with cancer	In depth history audit	Medical records of 18 children	The study found that physical aspects of the child's care were well documented. However 'human-to-human' contact was not well documented. The process of decisions to move from active attempts at cure to palliative therapy was not clear. The shift to palliative care was gradual rather then a definite move from cure to palliation. Relapse and disease progression were identified as important areas for further research.
Deasy-Spinetta, P	P Deasy-Spinetta. The school and the child with cancer. In: Living with childhood cancer, edited by JJ Spinetta and P Deasy-Spinetta, St Louis. Toronto. London:The C. V. Mosby Company, 1981, p. 153-168.	Teachers of children with cancer over three years	To investigate how children with cancer function as a group	Quantiiative	Development of a behavioural questionnaire over 3 years that was sent to teachers to complete about children with cancer and matched controls.	According to their teachers children with cancer where abscent more often then other children and struggled accademically. Teachers also suggested that children with cancer tended 'not to reach out to others' initiate activities or express their emotions.
Decker, C	C. Decker, C. R. Phillips, and J. E. Haase. Information needs of adolescents with cancer. J Pediatr Oncol.Nurs 21 (6):327-334, 2004.	74 adolescents newly diagnosed with cancer, and 39 adolescents with cancer who were 1-3 years from diagnosis	To describe how adolescents with cancer rated the importance of cancer related information.	Quantitative	Information Preferences of Adolescents Scale	Both samples scored highly for the need for information. Females had significantly higher scores in both groups. The young people added the following list of their personal information needs, treatment/side effects, uncertainty, social issues and personal/emotional issues.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Dixon-Woods, M	M. Dixon-Woods, C. Seale, B. Young, M. Findlay, and D. Heney. Representing childhood cancer: accounts from newspapers and parents. Sociol.Health Illn. 25 (2):143-164, 2003.	20 mothers and 3 fathers of children with cancer	To analyse newspaper accounts and parents accounts of childhood cancer	Qualitative	Interviews - narrative	It was found that newspaper accounts were eulogizing, constructing children as courageous, stoical, inspirational. Parents as 'battling the disease'. Parents accounts of children who are not uncomplaining or always cheerful. Parents experiencing role strain but unable to voice their situation.
Dolgin, M	M. J. Dolgin, S. Phipps, E. Harow, and L. K. Zeltzer. Parental management of fear in chronically ill and healthy children. J Pediatr Psychol. 15 (6):733-744, 1990.	31 children with chronic life-threatening illness. 30 children with chronic non-life threatening conditions. 28 healthy children.	To determine the impact of children's health status on parental management of fear and avoidance, and the relationship between the parents strategies and children's anxiety.	Quantitative	Child Development Questionnaire. Hospital Fears Rating Scale. State-Trait Anxiety Inventory for Children. Stait-Trait Anxiety Inventory. Primary Physician Ratings.	The study found that the chronically ill child's state of health (illness course, prognosis, physical impairment and time since diagnosis), rather than their diagnosis influenced parental behaviour. Parents of children who had a poor prognosis, less stability of illness course or greater physical impairment were said to foster more dependency. More anxious mothers tended to use less effective strategies in managing their children's fears about medical interventions.
Drake, R	R. Drake, J. Frost, and J. J. Collins. The symptoms of dying children. J Pain Symptom.Manage. 26 (1):594-603, 2003.	30 records of children who died. Interviews of 29 nurses regarding the child's symptoms in the last day of life. The nurses were interviewed within a week of the child's death.	To examine the symptom prevalence, characteristics and distress of children dying in a New Zealand Hospital in the last week of life.	Epidemiological	Investigation of medical records. Interview a nurse caring for each child in the last 24 hours of life. Each nurse also completed a revised Memorial Symptom Assessment Scale.	The average age of the children was 8.9 years. The dominant disease process was cancer and the most likely place of death was intensive care. 90% of the children had no pre-existing 'Do Not Resuscitate' order and this issue was addressed for the first time in the last days of life in 58% of the cases studied. The mean number of symptoms experienced by the children in the last week of life was 11.1 +/- 5.6. The records revealed a 'high burden of symptoms' in the sample studied and it is suggested that the application of the palliative care paradigm and a more proactive approach to symptom management might benefit dying children.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Earle, R	R. J. Earle, J. E. Rennick, F. A. Carnevale, and G. M. Davis. 'It's okay, it helps me to breathe': the experience of home ventilation from a child's perspective. J Child Health Care 10 (4):270-282, 2006.	Five children requiring home ventilation	To explore the experience of home ventilation from the child's perspective	Multiple case studies	Observations and audio taped interviews	Four themes emerged that were: 1. 'It's okay it helps me to breathe'. 2. The medicalization of childhood. 3. Being a child. 4. Hopes for the future. The children expressed acceptance of the ventilator and said they valued it because it helped them to breathe and feel comfortable. The children were knowledgeable and comfortable with the technology. The children were said to be much like other children and required ventilation at night – so that the technology did not intrude on other social settings like school. All of the children expressed hopes for their own future.
Eden, O	O. B. Eden, I. Black, G. A. MacKinlay, and A. E. Emery. Communication with parents of children with cancer. Palliat Med. 8 (2):105-114, 1994.	25 parents of children with cancer and leukemia	To assess the receptiveness of parents to information about their child's life threatening illness	Quantitative	Structured interview	23 sets of parents expressed 'deep shock and devastation' on hearing their child's diagnosis, 12 of these felt that they had taken in little or none of the information given. The parents were interviewed a few days after a long descussion with the consultant. The parents of 4 children denied that a long interview had taken place, 10 who remembered the discussion felt they did not understand specific details. All of the parents recounted a large number of talks with other professionals but some felt the information was confusing or conflicting. The majority of parents felt that their child had little or no understanding of 'what was going on'.
Glaser, B	BG Glaser and AL Strauss. Awareness of dying , Chicago:Aldine Publishing Company, 1965.	Adults and children dying in a hospital no clear description of numbers	To explore interactions between hospital staff and dying patients	Sociological study	Participant observation and interviews	It was found that the dying person and people caring for them maintained social order through four types of awareness; Open, closed, susepected and mutual pretence

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Gravelle, A	A. M. Gravelle. Caring for a child with a progressive illness during the complex chronic phase: parents' experience of facing adversity. J Adv Nurs 25 (4):738-745, 1997.	8 families (11 parents) of children with a progressive life-threatening illness, in Canada.	To explore the day-to-day experiences of parents caring at home for a child with a progressive life-threatening illness.	Qualitative	Phenomenological interview	The parents expereinces were conceptualised as an on-going process of facing adversity which included: planning/preparing, negotiating and utilizing resources. As parents had to continuously redefine and manage changes in their child's condition in the context on cumulative 'burden of care'. The many pressures on parental roles (especially for mothers) are considered.
Greenberg, L	L. W. Greenberg, L. S. Jewett, R. S. Gluck, L. A. Champion, S. L. Leikin, M. F. Altieri, and R. N. Lipnick. Giving information for a life-threatening diagnosis. Parents' and oncologists' perceptions. Am J Dis.Child 138 (7):649-653, 1984.	118 peadiatric oncologists and 31 families of children with acute lymphocytic leukemia (ALL).	To determine what information is percieved by parents and oncologists in the initial disclosure of life threatening diagnosis	Survey	Postal survey	Doctors and parents agreed on topics for the initial interview, diagnosis, prognosis, disease process, additional tests, therapeutic plan and the doctor's ability. The parents and doctors more or less agreed on the order of information to be given the doctors were more clear that parents should know that they might not retain all the information given at the first interview.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Grinyer, A	A. Grinyer and C. Thomas. Young adults with cancer: the effect of the illness on parents and families. Int J Palliat Nurs 7 (4):162-170, 2001.	22 parents of young people (aged 18-25 years) with cancer, all but five of whom had died	To examine how parents of young people with cancer describe their expereinces and those of their child	Qualitative	Independent narrative by parents, either written or taped.	Parents accouns suggested that young people struggled greatly with the loss of new found independence. Sometimes they made decisions against the wishes of their parents. Caught between the transition from school to adult life the young people did not have work colleagues or the stability of working relationships, whilst many of their old friends were moving on and away. Retaining a 'normal' life become complicated and still important. Sexual relationships and experience was discussed by some parents with their child, and they might not have before. Parents talked about their other children and explained that often friends didn't offer any practical help with teenage siblings. Financial pressures were mentioned, in one case the young person was spending lots on alternative therapy and research to find a treatment/cure
Haase, J	J. E. Haase and M. Rostad. Experiences of completing cancer therapy: children's perspectives. Oncol.Nurs Forum 21 (9):1483-1492, 1994.	7 children with cancer aged 5 -18 years, who had completed cancer treatment between 2 and 12 months before the study	To explore the child's perspective of completion of cancer treatment	Descriptive phenomenology	Open-ended qualitative interview	Six themed categories were described, realization of normal, hierarchical and cyclical recurrence fears, completions embedded in cancer exp, seeking a new normal, modifying relationships, resolution and moving on. The authors generated an 'essential strucure' from these 6 themes indicating that the expereince of completing cancer treatment had two faces from the children's point of view. One was celebration and hope the other was uncertainty and fear.
Hannan, J	J. Hannan and F. Gibson. Advanced cancer in children: how parents decide on final place of care for their dying child. Int J Palliat Nurs 11 (6):284-291, 2005.	Parents of five children who had died of cancer	To retrospectively explore the decisions made by parents about place of care for children dying of advanced cancer	Cross-sectional descriptive study	Semi-structured interviews.	This paper describes four of seven themes: valuing time left, needing to feel safe and secure, we didn't know what to expect, the difference between specialists and non-specialists. Parents valued time with their child to the end of the child's life and made decisions about the best place for care of the child as apposed to selecting a place of death.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Hechter, S	S. Hechter, M. Poggenpoel, and C. Myburgh. Life stories of families with a terminally ill child. Curationis. 24 (2):54-61, 2001.	4 families of children with terminal illness	To explore and describe the experiences of families with a terminally ill child and to develop an approach to facilitate families to obtain acquiescence.	Qualitative	Phenomenology. Multiple case studies	The researcher argued that family values changed when their child was terminally ill. Acceptance maked life easier. Living every moment to the full improved quality of life. Accepting support improved quality of life.
Hongo, T	T. Hongo, C. Watanabe, S. Okada, N. Inoue, S. Yajima, Y. Fujii, and T. Ohzeki. Analysis of the circumstances at the end of life in children with cancer: symptoms, suffering and acceptance. Pediatr Int 45 (1):60-64, 2003.	28 records of children who died of cancer, in the Hamamatsu University Hospital, Japan, between 1994-2000.	To analyze the signs and symptoms at the end of life in children with cancer	Quantitative	Medical records of 28 children	Symptoms included poor appetite 100%, dyspnea 82%, pain 75%, fatigue 71%, nausea/vomiting 57%, constipation 46% and diarrhea 21%. Results indicated that a large proportion of the children experienced symptoms that were not adequately comforted by palliative therapies. It was noted that no records showed whether children had spoken about their own acceptance, fear and awareness that they were dying.
Horstman, M	M. Horstman and A. Bradding. Helping children speak up in the health service. Eur J Oncol.Nurs 6 (2):75-84, 2002.	50 chronically sick children who mostly suffered from cancer, aged 6-10 years old and a group of 49 healthy school children	To investigate children's perceptions of health care professional, the environment and their need for information when sick	Qualitative	The draw and write technique and semi-structured interviews.	Overall the children wanted professionals to communicate with them directly about their diagnosis, treatment and prognosis. The children were able to describe how they percieved professional roles like nurses and doctors and their different expectations of each professional group. Humour was highlighted as a central skill for professionals communicating with children suffering from chronic illness.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Ireland, L	L. M. Ireland. Children's perceptions of asthma: establishing normality. Br J Nurs 6 (18):1059-1064, 1997.	10 children aged 9 -12 years with asthma.	To identify the beliefs and attitudes of asthmatic children regarding their illness and it's management.	Grounded theory	Open-ended qualitative interview	Core variable said to be 'establishing normality'. 4 major categories 'normal for me', 'paying attention', 'leaving asthma behind', 'discontinuity'. Discontinuity - feeling different to other children. Paying attention - the child's planning or preventative/problem solving behaviours. Leaving asthma behind - distraction, ignoring etc. Establishing normality - the primary focus of the children's management behaviour, used as a reference point to give meaning to the experiences. It was found that the children's concern to establish normality had some positive aspects but also lead the children to accept sub optimal control of their illness in certain circumstances.
Jennings, P	P Jennings. Coping mechanisms. Paediatric Nursing 4 (8):13-15, 1992.	Families of children with a life-threatening illness. 53 mothers in survey. 5 of these interviewed plus one professional per family.	To investigate how mothers cope at home when their child has a life threatening illness.	Quantitative and qualitative methods	Questionnaire to 53. Interview of 5 from the group + interview of one professional per these 5. Coping check list derived from empirical research (listing 22 coping strategies) were sent to the mothers	Mothers identified that they coped by, taking things day by day, crying, screaming, slamming doors, religion, prayer, religious groups, trying to forget, seeking knowledge, keeping occupied and hiding feelings. Professionals were unlikely to identify the strong emotional reactions expressed by the mothers.
Kreicbergs, U	U. Kreicbergs, U. Valdimarsdottir, E. Onelov, J. I. Henter, and G. Steineck. Talking about death with children who have severe malignant disease. N.Engl.J Med. 351 (12):1175-1186, 2004.	449 parents of children who died of cancer in Sweden	To investigate whether parents of children who are terminally ill talk about death with their child	Survey	Questionnaire	Most of the parents talked with each other about the child's impending death (76%), 27% talked with their child about death during the last month of the child's life. 25% used the word death. 46% of the parents reported that they did not sense that their child was aware that they were dying. Most parents who did sense that their child was aware of their own dying indicated that this occurred in the last month of the child's life, only 12% said this came up to 3 months before the child died. Mothers tended to spend more time with their sick child then the fathers and were more likely to express regret about not talking to the child about death. None of the parents who discussed dying with their child had any regrets.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Levealahti, H	H. Levealahti, C. Tishelman, and J. Ohlen. Framing the onset of lung cancer biographically: narratives of continuity and disruption. Psychooncology., 2006.	37 adult patients with lung cancer	To explore how people with inoperable lung cancer conceptualize their experience	Qualitative	Analysis of a subset of data from audio taped qualitative interviews from a larger study.	It was found that in addition to 'biographcal disruption' the study highlighted an alternative or parallel process which they called 'biogrphical continuity'.
Madden, S	S. J. Madden, S. E. Ledermann, M. Guerrero-Blanco, M. Bruce, and R. S. Trompeter. Cognitive and psychosocial outcome of infants dialysed in infancy. Child Care Health Dev. 29 (1):55-61, 2003.	16 children with end-stage renal disease undergoing long-term peritoneal dialysis. The ages of the children ranged from 1 to 12 years	To understand cognitive and psychosocial outcome of children with end-stage renal disease undergoing long-term peritoneal dialysis	A quantitative study using validated and standardized psychometric assessments	Measures of cognitive development: The Griffiths Mental Development Scales for children (children 6 years and under). The Wechsler Intelligence Scale for Children III UK (children older than 7 years through to 17 years). Psychological adjustment was measured using the Strengths and Difficulties Questionnaire. Impact on family life style was infered by the number and length of hospital admissions for the child in their first year of life.	67% of the children's scores fell within the average range. 50% of the children's psychosocial adjustment scores fell within the borderline to abnormal category, suggesting that the frequency of psychological difficulties was above the normal population. The authors suggested that the results illustrated how children undergoing peritoneal dialysis are faring better developmentally then they used to.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Mastroyannopoulou, K	K. Mastroyannopoulou, P. Stallard, M. Lewis, and S. Lenton. The impact of childhood non-malignant life-threatening illness on parents: gender differences and predictors of parental adjustment. J Child Psychol.Psychiatry 38 (7):823-829, 1997.	93 mothers and 78 fathers from families of children with non-malignant life threatening illness.	To describe families of children with life threatening illness and to explore gender difference between parents. Also to investigate the impact of the disease on family functioning and parental adjustment	Quantitative	Structured interview. The General Health Questionnaire. The Family Relationship Index. Nursing asessment questionnaire.	Mothers coped through emotional release. Fathers coped by withdrawal. Families wanted more information especially at times of crisis and felt isolated. Adjustment difficulties were greater nearer to the time of diagnosis. The child's health on one day was less significant then the overall prognosis and illness pattern on parental adjustment.
McGrath, P	P. McGrath. Treatment for childhood acute lymphoblastic leukemia: the fathers' perspective. Aust Health Rev. 24 (2):135-142, 2001.	4 fathers of children under treatment for Acute Lymphoblastic Leukemia (ALL)	To explore fathers' perspectives of caring for their child with acute lymphoblastic leukemia.	Part of a longitudinal qualitative study	Reflective open-ended qualitative interview	Highlights how the fathers experienced shock on hearing the diagnosis and emotional pain during the child's illness. The participants talked about the expression of pain through tears the researcher challenged steotypical images that 'men don't cry'.
Mullis, R	R. L. Mullis, A. K. Mullis, and N. F. Kerchoff. The effect of leukemia and its treatment on self-esteem of school-age children. Matern.Child Nurs J 20 (3-4):155-165, 1992.	13 children with leukemia (aged 6 -11 years) and 50 school age children without chronic illness	To investigate the self-esteem of school age children with leukemia and to compare this to the self-esteem of healthy children.	Quantitative	Kinetic Family Drawing-Revised and The Coopersmith Self Esteem Inventory	Children with Leukemia had lower self esteem then the healthy control group in terms of their school and academic performances. It is argued therfore that children with a chronic life threatening illness need transition support following their diagnosis and treatment.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Olausson, B	B. Olausson, Y. Utbult, S. Hansson, M. Krantz, M. Brydolf, B. Lindstrom, and D. Holmgren. Transplanted children's experiences of daily living: children's narratives about their lives following transplantation. Pediatr Transplant. 10 (5):575-585, 2006.	18 children and adolescents, aged 4-18 years.	To illuminate the meaning of transplanted children's experiences of daily living	Qualitative – phenomenology	Unstructured interviews	Two main themes emerged as: 1. Being satisfied with life – which included, being able to live a normal life, someone who cares and coping with one's new life. 2. Being dissatisfied with life – which included not being able to live a normal life, lacking someone who cares, not being respected and existential thoughts. Most of the participants were said to feel they led a 'normal' life and the others strived to achieve a 'normal' life. Social and mental support was important and had negative consequences if they were felt to be lacking.
Pendleton, S	Sara M. Pendleton, Kristina S. Cavalli, Kenneth I. Pargament, and Samya Z. Nasr. Religious/Spiritual Coping in Childhood Cystic Fibrosis: A Qualitative Study. Pediatrics 109 (1):e8, 2002.	23 children with cystic fibrosis aged 5-12 years.	To understand the role of religious/spiritual coping in children with cystic fibrosis.	Focused ethnography	Semi-structured interviews. The children were asked to draw God and themselves when they are sick. The children were asked about placement of components and to explain the meaning of their drawing. Parents were given a questionnaire designed for the study asking about their views of religious/spiritual behaviors in the family.	An overall domain of religious/spiritual coping emerged, divided into 11 themes: declarative, petitionary and collaborative religious/spiritual coping, belief in God's support, belief in God's intervention, belief that God is irrelevant, spiritual social support, ritual response, benevolent religious/spiritual reappraisal, punishing religious/spiritual reappraisal and discontent with God or congregation. The children who took part reported a range of religious/spiritual coping strategies for dealing with their cystic fibrosis.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Robinson, W	W. M. Robinson, S. Ravilly, C. Berde, and M. E. Wohl. End-of-life care in cystic fibrosis. Pediatrics 100 (2 Pt 1):205-209, 1997.	A sample of 44 records of children who died of cystic fibrosis in a hospital in Boston, 1984-1993	To examine differences in end of life clinical care for cystic fibrosis compared with terminal care in childhood cancer.	Quantitative	study of 44 medical records	The authors found that children who died of cystic fibrosis received a combination of palliative, therapeutic and preventative care to the end of their lives. The differences between the care found in this study and palliative care of children with cancer are highlighted. It is argued that therapeutic and preventative treatments for cystic fibrosis have a low morbidity and therefore there is more willingness amongst all involved to continue even if the outcome is uncertain. In fact they argue that the short-term fluctuations in respiratory function and uncertainly of immediate prognosis make it difficult for the patients, their family and medical teams to abandon therapeutic measures.
Rollins, J	J. A. Rollins. Childhood cancer: siblings draw and tell. Pediatr Nurs 16 (1):21-27, 1990.	20 healthy siblings aged 3-11 years of children with cancer.	To gain some understanding of siblings of children with cancer through analyzing drawings of their families.	Case study approach	Kinetic Family Drawing-Revised	The children used the drawing and talking to express 'non-disease' related stresses and concerns within the family. In one case the child expressed his isolation in a family barely communicating with each other and the important place of his brother for him. One of the siblings expressed concerns and fears about his brothers treatments. The authors discussed how siblings can be a great source of comfort to each other and highlight the need for professionals to be aware of the fears siblings face about loosing such relationships.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Saiki-Craighill, S	S. Saiki-Craighill. The personal development of mothers of terminal cancer patients: how Japanese women change through the experience of caring for and losing their children to cancer. Qual Health Res 12 (6):769-779, 2002.	57 mothers of children with cancer	To explore the experiences of bereaved Janpanese mothers of caring for their child who died of cancer	Qualitative	Open-ended qualitative interview	The mothers took on the role of primary care giver for social reasons, personal reasons or not being able to rely on others. They talked about fathers having to work, finding their own identity through this role, and being vital. The mothers expressed feelings of guilt and asked, 'why us?' Other issues included; learning from the child, being stong with the child not just for them; and difficulties of witnessing the deaths of other children in hospital from the same condition. They were found to be redefining themselves after the child's diagnosis and death - never being the same again. Living with the child in mind, hope to see them again, and 'trying to be what the child would have wanted me to'. ...
Samaniego, L	L. R. Samaniego, H. S. Caldwell, R. Nitschke, and G. B. Humphrey. Exploring the physically ill child's self-perceptions and the mother's perceptions of her child's needs: insights gained from the FIRO-BC--a behaviour test for use with children. Clin Pediatr (Phila) 16 (2):154-159, 1977.	Children aged 10-14 years with a life-threatening illness, children with a non-life threatening illness, and healthy controls. 21 children (7 in each group) and their mothers	To investigate mother's awareness of her ill child's needs and the dying child's needs and desires compared with children with non-life threatening illness and well children.	Quantitative	The Fundamental Interpersonal Relations Orientation Behaviour Test for Children (FIRO-BC)	The greater the severity of the child's illness the more the child reported 'wanting affection' this was said to be related to the child's desire to be appreciated, valued, as a result of the threat to their life. The children were however unlikely to voice this need, which might then go unexpressed and unaddressed. Children with non-life threatening illness wanted control of their illness and treatment. It is suggested that this might have been a reaction to the restrictions imposed on their developing need for independence from parents.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Snethen, J	J. A. Snethen, M. E. Broome, J. Bartels, and B. A. Warady. Adolescents' perception of living with end stage renal disease. Pediatr Nurs 27 (2):159-7, 2001.	35 adolescents with end stage renal disease, who were on renal dialysis or had received a renal transplant. The young people were aged 13 to 18 years.	To explore the perceptions of adolescents with end stage renal disease.	Q - methodology	Q method - The researcher and a panel of 'experts' developed a set of 48 statements from interviews with 15 adolescents with end stage renal disease not participating in this study. The adolescents sorted the statements into 'most like me', 'most unlike me', or 'has no meaning to me'.	4 perspectives held by the young people were a. normalization, b. illness intrusion: barrier to normalcy, c. illness management: parent focused. D. Illness management: self-focused. It is suggested that the young participants held more positive perspectives of living with end stage renal disease then previously described in the literature.
Spinetta, J	J. J. Spinetta, HH McLaren, RW Fox, and SN Sparta. The Kinetic Family Drawing in childhood cancer: a revised application of an age-independent measure. In: Living with childhood cancer, edited by JJ Spinetta and P Deasy-Spinetta, St Louis. Toronto. London:The C.V. Mosby Company, 1981, p. 86-120.	594 drawings (254 by patients, 154 by siblings, 135 by mothers and 51 by fathers)	To explore whether children's drawings can be used in research to explore emotional and attitudinal reactions to cancer	Kinetic Family Drawing	Kinetic Family Drawing	The authors argued for rigour and use of carefully structured scoring systems for analysis of children's drawings and cautions against over interpretation of children's emotions or feelings in their drawings.
Steele, R	Rose. G. Steele. Trajectory of certain death at an unknown time: children with neurodegenerative life-threatening illnesses. Can.J.Nurs.Res. 32 (3):49-67, 2000.	Families of children with neurodegenerative life threatening illness 8 families (29 family members, including observation of 10 sick children) Child participants - observed	To explore the experience of families caring for a child with a neurodegenerative life threatening illness dying at home.	Grounded theory	Interviews based on the grounded theory method	The study revealed that families were navigating uncharted territory, plateaus of relative stability and feelings of dropping off the plateau as the child dies. Entering unfamiliar territory. Shifting priorities. Creating meaning. Holding the fort.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Steele, R	R. G. Steele. Experiences of families in which a child has a prolonged terminal illness: modifying factors. Int J Palliat Nurs 8 (9):418-434, 2002.	Families of children with neurodegenerative life threatening illness. 8 families (29 family members, including observation of 10 sick children). The children's diagnoses were specifically not given to ensure anonimity.	To explore the experiences of families caring at home for children with neurodegenerative life threatening illnesses who were dying.	Grounded theory	Observation and interviews based on the grounded theory method.	Families were said to move through a process of 'navigating uncharted terrotory' in caring for their dying child at home. The strategies used by families were influenced by relationships with professionals, availability of information, gender differences, communication between parents.
Stewart, D	D. A. Stewart, A. Stein, G. C. Forrest, and D. M. Clark. Psychosocial adjustment in siblings of children with chronic life-threatening illness: a research note. J Child Psychol.Psychiatry 33 (4):779-784, 1992.	10 siblings (aged 6-17 years) of children using Helen House (a children's hospice in Oxford). Their parents. Also a matched group of healthy controls.	To carry out a controlled investigation of the psychological functioning and family relationships of the healthy siblings of children with chronic life-threatening illness	Quantitative	Siblings: -Piers Harris Self Concept Scale, Spielberger Anxiety Rating Scale. Birleson Depressive Rating Scale. Bene Anthony Family Relations Test. Semi-structured interviews. Parents: - The General Health Questionnaire, The Dyadic Adjustment Scale, The Social Adjustment Scale and the Rutter A2 questionnaire	There were no differences between the siblings and the healthy control group on measures of depression, anxiety and self concept. Siblings expressed more negative emotions in relation to their fathers, mothers being less involved in social activities and fathers being less involved with extend family members. 6 of the 10 siblings participating were very involved with caring for the ill child. 5 of the siblings had an understanding of their brother or sisters diagnosis and the other 5 had very little information and had hardly discussed it with anyone. 8 of the siblings felt the hopsice served a useful role for the family.
Stewart, J	J. L. Stewart. "Getting used to it": children finding the ordinary and routine in the uncertain context of cancer. Qual Health Res 13 (3):394-407, 2003.	11 children undergoing treatment for cancer. The children were aged 9-12 years.	To study children's reactions to uncertainty in the context of having cancer because uncertainty has been described as a major stressor faced by children with cancer.	Grounded theory	Semi-structured interviews	Children facing cancer treatment identified multiple uncertainties in their experience but did not characterize themselves as uncertain. The results indicated that the children quickly came to view their lives as routine and ordinary even though they were aware that the disease and it's treatment had many unpredictable aspects.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Straker, G	G. Straker and M. Kuttner. Psychological compensation in the individual with life-threatening illness: A study of adolescents with cystic fibrosis. S.Afr.Med.J 57 (2):61-62, 1980.	10 adolescents with cystic fibrosis, who were aged 12 -16 years. A control group matched for age, sex, religious denominations and home language.	To reinvestigate psychological functioning in adolescents with a chronic life threatening illness (cystic fibrosis).	Quantitative matched control study	Norwicki-Strickland Locus of Control test to assess the individuals perceptions about how much they had control over their lives. Thematic Aptitude Test was combined with a structured interview to assess peer and family relationships, anxiety about death and the future, depression and feelings for persecution.	The results demonstrated that there were no significant difference between the groups in psychological function. The authors argued for future studies to explore the ways in which adolescents with life threatening illness develop coping mechanisms.
The, A	A The. Palliative care and communication: experiences in the clinic, Buckingham. Philadelphia:Open University Press, 2002.	15 patients with lung cancer	To study the communication expereince of patients with small cell lung cancer	Ethnographic	Ethnographic study looking 30 patients wih small cell lung cancer.	Found that patients and clinicians focused on the short-term treatment options and 'hope' rather than the poor prognosis and likely illness trajectory of small cell lung cancer.
Thompson, R	R. J. Thompson, Jr., J. L. Zeman, D. Fanurik, and M. Sirotkin-Roses. The role of parent stress and coping and family functioning in parent and child adjustment to Duchenne muscular dystrophy. J Clin Psychol. 48 (1):11-19, 1992.	35 parents of boys (aged 4-14 years) with Duchennne Muscular Dystrophy	To explore stress and coping in parents of children with duchenne muscular dystrophy	Quantitative	Structured interview. Self-report questionnaire.	Just over half the parents reported high stress levels, high use of palliative rather then adaptive coping and poor adjustment were associated with family conflict and reports of behaviour problems for the children.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Tozer, R	R Tozer. At the double: supporting families with two or more severely disabled children, London:National Children's Bureau for the Joseph Rowntree Foundation, 1999.	24 families with two or more children with severe disabilities	To explore the whole families experience and support needs when caring for two or more severly disabled children	Qualitative	Interviews and observations in the family home. Interviews with key professionals nominated by families.	Families wanted professionals to understand what family life was like on a daily basis when caring for two or more severely disabled children. All family relationships were affected by the extra needs for support and requirement for at least two carers. This was reflected in unemployment of both parents and low family income. Siblings and grandparents were often involved with caring. Families most wanted professionals to see the children as individuals and have care co-ordinated by one or two trusted professionals.
van Veldhuizen, A	AM van Veldhuizen and BF Last. Children with cancer: communication and emotions, Amsterdam. Lisse.:Swets & Zeitlinger, 1991.	82 children, aged 4-16 years who were suffering from cancer and their parents	To investigate the impact of open communication between parents and their children with cancer	A mixed design using quantitative and qualitative methods	Structured interviews. Self report questionnaire. For children aged 4-7 years the researchers used a model of a hospital room and a set of dolls.	Found that open communication was multidimensional concept. Children informed about the diagnosis early on by parents experienced less anxiety or depression. Attempts by parents to control the flow of information were often futile and the children found out from 'blunt' peers or siblings. Children tended to loose confidence and assume the 'worst' when not informed. Among fathers more negative emotions were linked to open communication (they found it harder?). Many parents never asked about the child's emotions. Children wanted to control the frequency and content of discussions about the illness with parents, themselves. Communication aimed to reduce uncertainty and increase control. The child needed to believe the parents were 'strong' and the parents needed to believe the child 'could handle the situation and would survive'. 'Double protection'.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Walker, A	A. M. Walker, G. Harris, A. Baker, D. Kelly, and J. Houghton. Post-traumatic stress responses following liver transplantation in older children. J Child Psychol.Psychiatry 40 (3):363-374, 1999.	18 children post liver transplant, 18 children with asthma (chronic illness), 18 who had undergone non-life threatening surgery. The children were aged between 7 and 16 years.	To assess the psychological adaptation of children who have undergone a liver transplantation. To understand whether a liver transplant can be understood as a traumatic medical event sufficient to produce symptoms of post traumatic stress.	Case control	Interview using the Child Post-Traumatic Stress Reaction Index	The scores revealed a significant difference between the group of children who had undergone liver transplant and those who had a chronic life-threatening illness or those who had undergone routine surgery. It was found that the acute life threat involved in the liver transplantation contributed to the development of post traumatic stress symptoms in the severe range (11%), in the moderate range (11%) and in the mild range (56%). The authors argued that the combination of acute life threat (as a result of the liver transplant) and chronic life threat (from the child's underlying diagnosis) lead to the experience of more symptoms of post traumatic stress then if the children were exposed to either of these life-threats alone.
While, A	A While, C Citrone, and J Cornish. Bereaved parent's views of caring for a child with a life-limiting incurable disorder. London:King's College. 1996. Christopher's.	44 bereaved families	To explore the experiences of bereaved families whose chidlren did not have cancer.	Qualitative	Unstructured interviews with a topic guide used as a prompt	Parents described what they valued most about certain professionals, being able to ask for help. Critical of services that focused only on the child and not the whole family. Asking even for a couple of hours respite to take a bath, they identify the skills required as possibly coming from 'another mum'. Nobody asked them what they needed. Action depended on money. Sometimes equipment provided to help was useless and in the way. Poor advice about benefits. Suggested that written information to dip in and out of as they felt able would be helpful. They suggested that parents needed staying power to get what the child needed. Respite services where not helpful when parents had to come and provide the care themselves. Nothing for young adults. Only answering the family questions can cause problems if they don't know what to ask.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
While, A	A While, C Citrone, and J Cornish. A study of the needs and provisions for families caring for children with life-limiting incurable disorders. London:King's College. 1996. Christopher's.	99 families of children with life limiting incurable disorders	To provide evidence of the needs of families caring for a child with a life-limiting incurable disorder the palliative and terminal phases of the illness and to describe the extent and range of support services available to these families	A complex design of five elements including qualitative and quantitative aspects.	Analysis of mortality/morbidity data for England and Wales for 1987-1991. Interview of 99 families using a modified OPCS Disability Survey Interview Schedule. A postal survey of 74 statutory service providers in 4 Regional Health Authorities. Semi-structured interviews with 92 statutory service providers in 12 DHA's.	Analysis of childhood mortality indicated that 1,100 children die from life limiting incuarbale disorders in England and Wales per annum. Among the 99 families interviewed the most frequently reported disabilities related to locomotion, behaviour, continence and dexterity. Over half of the children were severly disabled and the severity of their disabilities increased with age. In addititon two thirds of the children required time consuming therapeutic regimes conducted by parents. The child's diagnosis and treatment had far reaching consequences for all the families. The families described many unmet needs. The survey of statutory provision revealed differences across health authority regions and an 'enormous variety of voluntary providers were found to be offering support to life-limited children and their families.
Wise, B	B. V. Wise. In their own words: the lived experience of paediatric liver transplantation. Qual Health Res 12 (1):74-90, 2002.	9 children, aged 7-15, who had undergone liver transplant	To uncover the experience of paediatric liver tranplant recipients from the period prior to transplant, through surgery and beyond.	Phenomonolgical	Phenomonology style interview with child to draw and talk	The following themes were suggested: the body 'upstaging' the developing self, a desire to fit in with peers. Physical signs of illness and being teased/supported by peers. Seeing problems that others faced. Understanding why other people don't get it. Thinking of the donor, but getting on. Being surrounded by sick children in hospital is difficult. Holding in and letting out pain/ medical procedures. Protecting parents/mother from every detail/sign of the illness. Holding on to the normal things/processes of childhood, barriers to normal from illness. The authors suggested that the children faced many challenges including surviving a life threatening illness and normal developmental tasks and the major theme related to the children striving for normalcy in their lives.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Wolfe, J	J. Wolfe, N. Klar, H. E. Grier, J. Duncan, S. Salem-Schatz, E. J. Emanuel, and J. C. Weeks. Understanding of prognosis among parents of children who died of cancer: impact on treatment goals and integration of palliative care. JAMA 284 (19):2469-2475, 2000.	103 parents of children with cancer. 42 pediatric oncologists	To examine understanding of prognosis among parents of children with cancer	Quantitative	Survey of parents and pediatric oncologists. All items closed with categorical responses or Likert scale.	The results suggested that the doctors recognized that the children had no hope of cure before parents. Earlier recognition by parents and doctors was associated with stronger emphasis on treatment directed at lessening suffering.
Wolfe, J	J. Wolfe, H. E. Grier, N. Klar, S. B. Levin, J. M. Ellenbogen, S. Salem-Schatz, E. J. Emanuel, and J. C. Weeks. Symptoms and suffering at the end of life in children with cancer. N.Engl.J Med. 342 (5):326-333, 2000.	103 parents of children who died of cancer, between 1990-1997 in a Boston hospital	To investigated how bereaved families of children with cancer percieved their child's symptoms and suffering at the end of life.	Qualitative	Semi-structured interviews.	Parents reported that their children had received 'aggressive treatments' to the end of life and that palliation of symptoms like pain, fatigue and dyspneea were inadequate in 89% of the cases. It is suggested that children dying of cancer have substantial suffering in the last month of life and attempts to control their symptoms are not always successful.
Woodgate, R	R. L. Woodgate and L. F. Degner. Cancer symptom transition periods of children and families. J Adv Nurs 46 (4):358-368, 2004.	Families of children, aged 4-18 years, with oncology diagnosis. 39 children and their families including siblings and parents	To explore and describe the symptom course in childhood cancer as expereinced by children and their families.	A longitudinal qualitative study from the interpretive interactions perspective	Open-ended qualitative interview. Participant observation.	The author proposed that interrelationships existed between the child's symptoms of cancer and the 'families way of being in the world'. The more the symptoms intruded on family life the harder it is to maintain daily life and live with the notion of a healthy child with cancer. The child's symptoms therefore had an impact on all aspects of the families daily life.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Woodgate, R	R. L. Woodgate and L. F. Degner. A substantive theory of Keeping the Spirit Alive: the Spirit Within children with cancer and their families. J Pediatr Oncol.Nurs 20 (3):103-119, 2003.	Families of children aged 4-18 years with oncology diagnosis. 39 children and their families including parents and siblings	To understand the experience of childhood cancer from the child and family's perspective	A longitudinal qualitative study from the interpretive interactionist perspective	Open-ended qualitative interview. Participant observation.	The study provided an understanding of how children and families used the process of 'keeping the spirit alive' in the context of the child's cancer diagnosis. The central phenomenon identified as having the biggest impact on the children and their families was 'getting through all the rough spots'. It was found that support from within the family was key to maintaining each person's sense of spirit and keeping the family unit together.
Woolley, H	H. Woolley, A. Stein, G. C. Forrest, and J. D. Baum. Imparting the diagnosis of life threatening illness in children. BMJ 298 (6688):1623-1626, 1989.	25 families pilot. Main study 24 families just referred to the children's hospice, 21 families using the children's hospice (all mothers, 25 fathers present at interviews).	To explore how parents percieved the way they were told their child's life threatening diagnosis.	Quantitative and qualitative	Semi-structured interviews. General Health Questionnaire.	Parents valued "open, sympathetic, direct and uninterrupted discussion of the diagnosis in private that allowed sufficient time for them to take the news in and for doctors to repeat and clarify information". P1623.
Woolley, H	H. Woolley, A. Stein, G. C. Forrest, and J. D. Baum. Cornerstone care for families of children with life-threatening illness. Dev.Med.Child Neurol. 33 (3):216-224, 1991.	Families of children with progressive, life-threatening illness. (45 families) 24 families referred to a children's hospice but who had not yet used it. 21 not so far planning to use the children's hospice.	To examine the role of co-ordination of care for families of children with life theatening illness.	Quantitative and qualitative	Semi-structured questionnaire. Parents also explained their own experiences in more detail.	11 families had no corner stone carer, mostly these children had non-malignant disease. Parents valued from professionals. Mutual respect and compatibility, predictability and mutually agreed contact, acceptance of their distress, understanding the illness or honesty about the limits of their knowledge, knowledge of support and other benefits, continuity of care through life and bereavement, humour and the ability to remember the child together (in small ways).
Wrght, P	P. S. Wright. Parents' perceptions of their quality of life. J Pediatr Oncol.Nurs 10 (4):139-145, 1993.	55 parents of children with cancer	To investigate parents perceptions of their quality of life after their children were diagnosed with cancer.	Quantitative	Completion of Varrichio-Wrigth Impact of Cancer Questionnaire-Parents.	Parents were found to have a good quality of life but not as good as it was before their child was diagnosed. Discussion of the impact of being 4 years away from diagnosis and how the immediate threat had diminished.

First Author	Reference	Participants	Aims	Design	Methods	Key findings
Young, B	B. Young, M. Dixon-Woods, M. Findlay, and D. Heney. Parenting in a crisis: conceptualising mothers of children with cancer. Soc.Sci Med. 55 (10):1835-1847, 2002.	20 mothers of children with cancer	To investigate the experiences of mothers living with a child with cancer	Grounded theory	Semi-structured interviews.	The mothers experienced biographical disruption as soon as they noticed something was wrong with their child that was increased with diagnosis. It impacted on their sense of self, social identity and life roles. They faced new responsibilities including what the authors called 'an obligation of proximity' to the child; to be physically close to the child providing comfort and keeping watch. The life threat to their child put everyday roles and concerns under a new light.
Young, B	B. Young, M. Dixon-Woods, K. C. Windridge, and D. Heney. Managing communication with young people who have a potentially life threatening chronic illness: qualitative study of patients and parents. BMJ 326 (7384):305, 2003.	Families of children with potentially life threatening chronic illness. 13 mothers, 6 fathers, 13 young people	To examine young people's and parents' accounts of communication about cancer in childhood.	Qualitative	Qualitative interviews using topic guides. 8 young people were interviewed alone, 5 with their parents.	Disclosure of the diagnosis was to parents without the child in all but 2 cases. The children had mixed feeling about whether they wanted to have been present or not. Parents on the whole preferred to hear it first alone - to ask questions and not breakdown in front of the child. Parents and children described executive and control roles of parents and partnership roles of parents. Young people were aware that at times they were not being told everything that their parents were. At times they selected who to ask because that person would tell them the 'truth'. Discussion of young person's social position and questions about their, ability to comprehend. The children felt there were some things that they did not want, or need to know. Young persons age did not reflect what they needed, wanted re communication.
Zani, B	B Zani, A Di Palma, and C Vullo. Psychosocial aspects of chronic illness in adolescents with thalassaemia major. Journal of Adolescence 18:387-402, 1995.	Adolescents with thalassaemia major and healthy controls.	To evaluate the impact of thalassaemia major on the psychological functioning and social behaviour of adolescents	Quantitative	A questionnaire developed for the study to gather sociodemographic characteristics. Adolescent Coping Orientation for Problem Experiences scale.	The adolescents with thalassaemia had normal psychological and social development and did better then their peers in scores for self-esteem, self description and functional coping strategies. It is suggested therefore that chronic illness does not necessarily imply negative psychogical and social development and may strengthen the resources of some young people.

TABLE OF DISCUSSION PAPERS

First Author	Reference	Summary of main points
Adams, D	DW Adams and EJ Deveau. Beyond the innocence of childhood: Factors influencing children and adolescents' perceptions and attitudes towards death, Amityville. New York:Baywood Publishing Company INC, 1995.	We discuss the process of birth with children but rarely the process of dying p9 Argues we hope that children will have little direct experience of death, don't want to say the wrong thing and upset/confuse them, believe that children under a certain age can not comprehend. But they will have questions, miss the person, deserve honesty and kindness, will feel loss and sadness and might have other questions as they develop.
Alderson, P	Priscilla Alderson and Christopher Goodey. Research with disabled children: how useful is child-centred ethics? Children & Society 10:106-116, 1996.	Paper discussing the place of disabled children in research and argues that the complications arise not from the child's disabilities but from the place that children are given in western society. Interesting deconstruction of researchers fears 'are they going to talk to me?'. Assumed difference and remoteness of children. Risk of accepting anything they say for fear of probing too deep or upsetting. Children's ability to set the agenda and control their participation. To ask why are you asking? Barriers, parents, teachers, words. How to work with the family during a group interview but still keep the focus on the child. Children with profound needs as a paradigm for involving all children.
Allard, A	Amanda Allard. Involving young people - empowerment of exploitation? Children & Society 10:165-167, 1996.	Short discussion re involving children in research and other policy/service developments. Suggests to avoid exploitation there must be a genuine desire to listen, children who want to participate, worker with the skill and courage to involve the children.
Atkinson, D	D Atkinson. Research interviews with people with mental handicaps. Mental Handicap Research 1 (1):75-90, 1988.	Paper about how to involve vulnerable people in research. Sensitive discussion of methods and what seemed to work. "It means investing time in personal contact and, within the context of a relationship, allowing a full picture of a person to emerge." p76 Aims like mine see p78. Nice discussion. Read again.
Barnard, P	P Barnard, I Morland, and J Nagy. Children, bereavement and trauma: nurturing resilience, London:Jessica Kingsley Publishers, 2003.	The authors describe their work with children bereaved after the Hillsborough disaster. They argue for uncritical listening to children and understanding the needs of bereaved children as different to the needs of adults. A short section provides insights from children about what helped them.
Beresford, B	B Beresford. Personal accounts: involving disabled children in research, London:The Stationery Office, 1997.	Argues for a move towards involving disabled children in research, seeking and valuing their opinions. The ethical and methodological issues are explored. Also examined are the issues related to involving children with communication or learning difficulties in research.
Cooley, C	C. Cooley, S. Adeodu, H. Aldred, S. Beesley, A. Leung, and L. Thacker. Paediatric palliative care: a lack of research-based evidence. Int J Palliat Nurs 6 (7):346-351, 2000.	Highlight limited literature on children needing palliative care, especially of non-malignant disease. Limited understanding re the child's concept of death when they are life-limited or terminally ill. Call for research with children rather than on them. Identifies ethically issues and consent.
Corr, C	C. A. Corr and D. M. Corr. What is pediatric hospice care? Child Health Care 17 (1):1-11, 1988.	To maximize present quality of living. Hospice care as a type of palliative care? Palliation is in the mainstream of medical care e.g. the common cold. Child as part of their family. Home care as desirable, to enable child and family to live in the 'customary setting of their lives'. Aim of home care 'an atmosphere of safety and co-ordination'. Advocacy role of hospice for these children and their families at personal and social levels. Advocacy at the social level via research.

First Author	Reference	Summary of main points
Corr, C	C. A. Corr and D. M. Corr. Children's hospice care. Death Stud. 16 (5):431-449, 1992.	Limited research highlighted, call for additional and more sophisticated research. Hospice as a philosophy not a facility. To help people live not die. Holistic support of the child and their family. P434 "And both patients and family members must be understood and helped to function as part of the team that is giving care. This is particularly important in situations involving children as one strives to empower parents to be the primary care givers for their children".
Crossley, D	D Crossley, M Lewis, S Lenton, P Stallard, and S Bailey. What is a life threatening illness? Working definitions in paediatric clinical practice a discussion of the issues. 1994.	Describe the changing picture, increasing survival due to advances in medical treatment. Conditions are said to be both a threat to life and limit to life expectancy. In care it is said to be best to take each case on an individual basis. A non-categorical approach was taken because diagnosis was rarely helpful in providing the child's prognosis. Key to consider each child on an individual basis.
Davis, J	JM Davis. Understanding the meanings of children: A reflexive process. Children & Society 12 (5):325-335, 1998.	What are children's voices in research and how should we listen to them. 1. One voice, one culture, different to adults 'The tribal child'; 2. Groups of children with common meanings and behaviour, individual voices heard but not explored in detail; 3. Third form of qualitative research concerned with culture, difference and the variety of children's childhoods. Risks of adult - child power relations, exploitation of the child and adults avoiding the difficult questions and trying not to upset children. Highlights the many methods and asks what works? Child can control participation with silence, humour, conflict and shutting the gates. The roles of the researcher are discussed and may be varied, why is she asking this question (from her life and experience). An excellent paper - go back to it.
Dixon-Woods M	M. Dixon-Woods, B. Young, and D. Heney. Partnerships with children. BMJ 319 (7212):778-780, 1999.	Paper asking what is the impact on the child of participation in research or decision making in services. Raises concerns about the impact on the child of participation. Calls for better understanding of methods for involving children with learning difficulties. Suggests that information especially about services is often directed at parents.
Doka, K	K Doka. Talking to children about illness. In: Children mourning, mourning children. The Hospice Foundation of America, 1995, p. 31-39.	Society tries to protect children from illness and death on one hand and bombards them with images, verse, play, games and stories on the same theme. Children have several sources of information that tell them what is going on; external clues, internal clues, environmental, social. Prof to ask themselves what does the child need to know? What does the child want to know? What does the child understand?
Easson, W	WM Easson. The dying child: The management of the child or adolescent who is dying, Springfield. Illinois:Charles C Thomas Publisher, 1970.	Suggests that one should define exactly what aspect of death/dying is being studied. E.g. is the study setting out to examine the child's understanding of death or the child's management of a fatal process? Maybe the response of a bereaved child to the death of another. The child might also be learning what of his own responses are socially acceptable, in keeping other people comfortable.
Farrant, A	A Farrant. Death and dying children in children's literature. In: Sibling bereavement: Helping children cope with loss,Anonymous London:Casssell, 1998, p. 4-8.	Highlights how graphic images on TV or sentimental images of dying children in children's literature, both avoid the pain of grief and loss for all involved. In children's literature 'only the good die young', 'death of the virtuous child - assured of a place in heaven' 'showy sentimentality to avoid the pain and grief of those left behind.

First Author	Reference	Summary of main points
Faulkner, K	K. W. Faulkner. Talking about death with a dying child. Am J Nurs 97 (6):64, 66, 68-64, 66, 69, 1997.	Uses one case of a dying 5 year old child to discuss communication with dying children. Outlines the classical approach of Piaget but states "Of course, most children with a life threatening illness have a greater understanding of death then other children of their age. And children with terminal disease are usually aware of their condition, though some go to great lengths to hide that knowledge from the adults around them." Offers a set of simple guidelines for adults caring for dying children.
Glaser, B	BG Glaser and A Strauss. Time for dying, Chicago:Aldine, 1968.	After the publication of 'Dying awareness' Glaser and Strauss suggested a link between dying and certainty and identified four death expectations; certain death at a known time, certain death at an unknown time, uncertain death but a known time when the question would be resolved, uncertain death and unknown time when the question would be resolves
Gold E	Elizabeth-Anne Gold. The role and need of the children's hospice in the United Kingdom. International Journal of palliative Nursing 3 (5):281-286, 1997.	Scarcity of empirical research. Limited literature. Suggests that children's hospices could be seen as 'local centres of clinical research, education and audit rather than just a provision of care'. Extended summary of the Helen House study. Suggests that 'all decisions by professionals must be taken with families'. Everyone including siblings should have a voice. Families should have choice of local or hospice respite. 'Drawback' of hospice environment with other terminally ill children, which some families may not find beneficial.
Goldman, A	A. Goldman. ABC of palliative care. Special problems of children. BMJ 316 (7124):49-52, 1998.	A paper developing the idea of palliative care within paediatrics as a speciality. A summary of the ACT paper. "Often a picture must be built up through discussion with the child" advocates offering children choice. Summary of the group of children, their needs, family needs and palliative care treatments.
Hilden, J	J. M. Hilden, J. Watterson, and J. Chrastek. Tell the children. J Clin Oncol. 18 (17):3193-3195, 2000.	A description of practice using two family stories. Firstly, a six year old girl whose portents do not want to discuss her poor prognosis with the child or her 8 year old sister. The sister expressed her awareness and fears about the death of her sister to care staff. The sister was sent to relatives as the child died and was said to have behaviour problems and look bewildered at the funeral. Secondly, an 8 year old boy the family took the opposite approach, trying to be open with the child and his sibling. The authors argue for greater support of families who have to communicate with their dying child and better training of professionals in a range of approaches. It is suggest that parents can gently observe how much their child seems to understand about their illness.

First Author	Reference	Summary of main points
Hockley, J	J. Hockley. Psychosocial aspects in palliative care--communicating with the patient and family. Acta Oncol. 39 (8):905-910, 2000.	Argued for professionals to be alert to 'unspoken fears' and 'conspiracy of silence' - active listening and non-verbal communication being described as key skills. Asks if the hospice movement really sees the needs of the family and patient as a unit or as two separate identities. How many family meetings? For professionals to consider what other stress' the family might be facing in their lives, work, adolescence, how long they have been managing the illness. Changing roles because of the illness and death. Case studies p907-908 including how children are affected. Warning children about an anticipated death is painful but important. Suggests that socially we still assume that children are too young to understand a great deal about death and dying. 'But children not given accurate information will often try to make up a story to fill the gaps of hide themselves away in a fantasy world' p909. Argues that honesty and showing emotion with children is not detrimental so long as the child has a sense of control in the situation for themselves and from the adult. Highlights how in the UK a Health Visitor would generally be asked to help with a child who is bereaved or facing the death of a loved one especially a parent. But argued that skill is more central then professional discipline.
Hynson, J	J. L. Hynson, J. Gillis, J. J. Collins, H. Irving, and S. J. Trethewie. The dying child: how is care different? Med.J Aust 179 (6 Suppl):S20-S22, 2003.	Description of paediatric palliative care and how children have special needs. Calls for attention to the child's stage of development. Differences between very young children who may fear separation and adolescents who may fear changes in body image as a result of the illness or treatment. The authors argue for integration of curative and palliative care in paediatrics especially where there is uncertainty about prognosis.
Jankovic, M	M. Jankovic, N. B. Loiacono, J. J. Spinetta, L. Riva, V. Conter, and G. Masera. Telling young children with leukemia their diagnosis: the flower garden as analogy. Pediatr Hematol.Oncol. 11 (1):75-81, 1994.	50 children aged 6-15 were entered into a programme where their Dr told them their diagnosis without parents present. The Dr used slides and the idea of a garden with flowers and weeds to explain leukaemia to the child. The child was then asked to explain their diagnosis to their parents to initiate family discussion. 4-5 days later the child was asked to explain their diagnosis again to a psychologist and nurse and again 4-5 months later. The children understanding compared with Piaget stages p79. A score was given for understanding from 3 complete to 0 very poor 45 scored 3 or 2. Those who scored only 1 had a mean age of 7 and were the youngest. All the children and families are said to have valued the process and the children for being informed directly about their illness.
Judd, D	D Judd. Give sorrow words: working with a dying child, London:Whurr Publishers Ltd, 1995.	The author described her own work with a dying child. She argued that children who come into close contact with others who die or who are dying themselves are likely to have a more developed concept of death then other children of their age. Judd argued that as adults if we are evasive and don't answer questions we can deprive children of the opportunity to talk about their fears. It is recognised that as adults the difficulty of facing a child's death with them can become more painful as the child deteriorates. Judd argued that all children are different and therefore adults can take cues about how to talk from the child's questions and reactions. Experience of the death of another child with the same condition is said to be pivotal.

First Author	Reference	Summary of main points
Kane, J	J. R. Kane and M. Primomo. Alleviating the suffering of seriously ill children. Am J Hosp.Palliat Care 18 (3):161-169, 2001.	Gives numbers of children dying in USA. Argues that serious disease 'threatens the child's sense of personal integrity' and that the experience of suffering as far ranging consequences for the child's life and family unit. The essential needs of human said to be 'a sense of completion in one's life' and a 'sense of meaning'. Strategies directed to improve and enhance communication between children and family members. Argues that it is futile and potentially harmful to keep information from children. To let the child's questions guide discussions. Use of stories, games and artwork. Suggest that children as young ad five know more about death and dying then the classical approach suggests - when they have a serious illness themselves. In their experience children are more open then adults. Argues for change in some USA hospice care policies for children.
Kroll, L	L Kroll and B Jacobs. Children coping with the death of a sibling. Journal of The Royal Society of Medicine 88 (August):426-427, 1995.	Review of research into the effects on siblings when a brother or sister dies. Highlighted that studies have found that death of a sibling can have a direct effect on children's behaviour and that parents suffering bereavement themselves may distort how they perceive the child's behaviour. Argued that professionals and parents should be aware that young children especially often show their distress through their behaviour rather then words.
Kubler-Ross, E	E Kubler-Ross. On children and death: How children and their parents can and do cope with death, New York:Simon & Schuster, 1983.	A discussion of the authors many years of work with dying children. She explored the verbal and non-verbal communication methods used by dying children and argued that children have an inner or spiritual awareness of their own dying.
Kunin, H	H. Kunin. Ethical issues in pediatric life-threatening illness: dilemmas of consent, assent, and communication. Ethics Behav. 7 (1):43-57, 1997.	A discussion of the ethical issues in paediatric life threatening illness. The child's role in the process and age appropriate information. The issue of autonomy for children under 18 - finding ways to involve them in the process and hear from their point of view. Argues that children as young as 9 have been able to express preferences re treatment and in that way take part in decisions. Discussion of the term 'assent'. However involving children has risks - they might want to veto a decision, and it is unfair to ask a child about a decision already made. Discusses parental fears of what to tell the children and how to keep hope alive, also that children and parents may have different priorities in terms of what information they want. Children's fears when no one talks. "It is not unusual for families to return to old patterns of communication and meaning structures during times of intense stress and the prospect of losing a child to an illness raises additions considerations". P54
Levetown, M	M. Levetown. New programs for children living with life-threatening conditions. Tex.Med. 97 (8):60-63, 2001.	Poor understanding of child's concept of death, child as only person with full understanding of their own suffering but rarely consulted. Decisions made based on hoped for benefits but not full consideration of potential harms.
Lewis, M	M Lewis and D Crossley. The Lifetime Project: An assessment and resource service for children with a life threatening illness and their families. 1994.	Paper describing the development of assessment tools for the lifetime project. Listening is described as the gap in service provision.
Lonowski, S	S. C. Lonowski. Recognizing the right of terminally-ill mature minors to refuse life-sustaining medical treatment: the need for legislative guidelines to give full effect to minors' expanded rights. J Fam Law 34 (2):421-445, 1995.	Reference to adults rights for bodily self-determination and to refuse life-sustaining treatment and how this right is rarely offered to mature minors. 'The right to be let alone'. Highlights the lack of guidance for courts to enable them to make decisions with consistency and fairness, to respond efficiently and effectively to the needs of terminally ill children and their families.

First Author	Reference	Summary of main points
Matthews, GB	GB Matthews. The philosophy of childhood, Cambridge. Massachusetts. London.:Harvard University Press, 1994.	A review of classical accounts about how children come to understand about death and dying. The author argued that because children with direct experiences of death and bereavement are rarely included in research looking at children's concepts of death, it is unclear whether classical accounts would apply to them.
Michael, B	B. E. Michael and D. R. Copeland. Psychosocial issues in childhood cancer. An ecological framework for research. Am J Pediatr Hematol.Oncol. 9 (1):73-83, 1987.	Analysis of gaps in research focus and methods and suggests little attention has been given to whole family, school and other social networks and recommends use of the ecological systems framework for further research of children with cancer. Even more rare are studies that look at the wider social networks and society in which the child does not participate but is still influenced by. Good literature review.
Mohan, A	Ann Mohan, Caroline Glendinning, Karen Clarke, and Gary Craig. Researching children: Methods and ethics. Children & Society 10:145-154, 1996.	Paper drawing on research experience with children. Talks about focus on the problematic, use of proxy and need for children's voices to be heard. Child's own unique experience. Child as protector of adults. Researcher - child power relationship said to be more difficult in one-to-one situations. How children decline; outright, missing appointments, loss of interest during interview.
Murray, S	S. A. Murray, M. Kendall, K. Boyd, and A. Sheikh. Illness trajectories and palliative care. BMJ 330 (7498):1007-1011, 2005.	A review of papers and data from previous studies about illness trajectory. Three different illness trajectories of people who would die in the (relatively) near future were identified. These were: 1. Short period of evident decline (e.g. cancer). 2. Long term limitations with intermittent serious episodes (e.g. heart failure). 3. Prolonged dwindling (e.g. Alzheimer's disease). The concept of trajectory was said to be a potential benefit to the dying person, family and support services in helping to plan and meet needs. The limitations of trajectory were identified as the potential for social changes or atypical disease processes experienced by individuals.
Oppenheim, C	C Oppenheim, C Wallace, and N Wallace. Children's health care: Brief report Having a bone marrow test: A child's perspective. Children's Health Care 12 (1):41-42, 1983.	Describes involving one child, Holly and her mother in the process of creating a slide show to tell children about having a bone marrow test. Holly had numerous such tests and was guided by medical staff in creating a script for the slide show, including what she saw, felt, the room, the staff etc. Holly is said to have decided to participate in every stage of the process of creating the slides and recording the script herself. She apparently gaining a sense of achievement for her role.
Patistea, E	E Patistea. Family life, two cultures and a serious childhood illness. Community Practitioner 73 (4):566-569, 2000.	Points out that Leukaemia was acute and fatal, but is now considered a chronic illness. Case assessment based on 'Friedman's Model' based on theoretical perspectives: a structural functional perspective, a family developmental perspective and a systems perspective. The family are described in terms of being "an open social system with boundaries, self regulatory mechanisms, and interacting sub-components." p567. Then via links to socio-ecological and environmental networks. Interesting for the theory used.

First Author	Reference	Summary of main points
Penson, R	R. T. Penson, P. K. Rauch, S. L. McAfee, B. J. Cashavelly, K. Clair-Hayes, C. Dahlin, K. M. Green, B. A. Chabner, and T. J. Lynch, Jr. Between parent and child: negotiating cancer treatment in adolescents. Oncologist. 7 (2):154-162, 2002.	Discusses the case of a 19 year old woman with acute leukaemia. The psychiatrist suggests that the young lady wanted people to 'know' her as her 'well-accomplished and independent self in counterpoint to the ill, dependent person in the hospital'. The social worker talks of the young person and her mother as well defended, proud and private. The word nurse talks of an adolescent who wanted to be independent and take control of her disease and treatment. When the patient's treatment failed and options for other treatment ran out - she would not talk for a time and then talked to the nurse about what it might be like to die, to go to heaven. To be an angel. She asked to nurse to infuse the whole dose of morphine and get it all over with. The Oncologist talks of the pain in telling a young person 'the truth' and how difficult they found her devastation, although understanding it. The patient wanted to die at home and the team facilitated that. The psychiatrist suggested that the 'cornerstone' of this patient's self-esteem was her maturity and independence the approach to communication is outlined see p159 respecting her image of her best self, the opportunity to discuss fears, feelings, thoughts about treatment, palliative care and death. Developmental approaches and adolescence outlined on p159 Piaget, Erickson - cancer described as a challenge to all of the self-esteem, self-development work of adolescence. The paper suggests that at a time when adolescents would generally be moving away from family and home life threatening illness drives them back and sometimes into isolation both socially and emotionally - conversely the young person may find new meanings about what is important in life and the things that others of their age take for granted. The team talk of encouraging the young people to 'help each other' altruism, solidarity and connectedness with each other and the staff, to 'fuel personal growth and healing'. (Sounds like resilience as described by
Schmidt, S	S Schmidt, C Peterson, and M Bullinger. Coping with chronic disease from the perpective of children and adolescents - a conceptual framework and its implications for participation. Child Care Health Dev. 29 (1):63-75, 2003.	"The notion of adaptive coping as used by this research group indicates that coping is more than a simply a strategy, it is a cumulative history of interactive processes that are embedded in developmental organization. Interesting conceptual framework that links with mine. A framework for the study of coping in chronic conditions for adolescents that has regard for development, arguing they are inherently connected. Summary of system for review of the literature good p64. Links non-categorical approaches to study of chronic illness in childhood, to look at the common dimensions that show a high variation between illnesses. Advocates that attachment theory should be utilized to explore the parent-child dyad as a buffer for the child during stressful events. The impact of the child's dependence on parents and perhaps technology limiting development of peer relationships.
Sheldon, F	F. Sheldon and P. Speck. Children's hospices: organizational and staff issues. Palliat Med. 16 (2):79-80, 2002.	Brief discussion of how little research exists in paediatric palliative care, esp. children's hospices. Compares the Helen House studies with the Rowntree work. Discussion of ACH 'fierce rebuttal' of Rowtree study is connected with levels of staff stress. Highlights how advances in medicine mean the children will live longer and 'sicker'. Call for research into staff stress.

First Author	Reference	Summary of main points
Sourkes, B	B. Sourkes. Views of the deepening shade: psychological aspects of life-threatening illness. Am J Hosp.Care 4 (3):22-29, 1987.	Discussion about the impact of life threatening illness on 'patients', families and care givers. Argues that time can no longer be taken for granted and is experienced differently - focus on the present framed by themes of separation and loss. Shatters the orderly, predictable unfolding of time. Explores the experience via a thematic framework of loss of control, loss of identity and loss of relationships. Time as the trajectory of the illness, diagnosis, neutral time (living-dying interval) when the illness becomes an integral force within the family and gives way to 'normal life' and daily living. Also elective cessation of treatment, relapse, and anticipatory grief as well as the 'terminal' phase.
Strauss, A	A Strauss. Dying trajectories, the organisation of work and expectations. In: Death, dying and bereavement, edited by D Dickenson, M Johnson, and J Samson, London. Thousand Oaks. New Delhi:The Open University Press, 2000, p. 196-199.	A discussion highlighting key points in the dying trajectory focusing on the last days or hours of life
Stuber, M	M. L. Stuber. Psychiatric sequelae in seriously ill children and their families. Psychiatr Clin North Am 19 (3):481-493, 1996.	Discusses changes in survival, intensive treatment and therefore the experience for the child and family is no longer one of 'certain death' but complex life threat over long periods of time (Chronic). Requiring adaptation by all family members. Causes of psychiatric sequelae, diagnosis, treatment, not just physical impairment but functional impairment, the child's perception of events and consequences (locus of control), disruption to the developing sense of identity through isolation, missed education and bodily disfigurement like scaring or other, social reactions (fears of catching something etc), the child's fears of being seen as different. Family members facing emotional reactions of the child and themselves, 'ethical concerns' and 'perplexing choices' re intensive treatment. Mediating factors, the child's perception of events and personal ability to cope, evaluation of personal threat influenced by their age, stage of development (refers to Piaget) and experiences. Social/family support. Discusses 'open communication' with the child and ideas related to not taking away a 'person's hope'.
Sutherland, R	R. Sutherland, J. Hearn, D. Baum, and S. Elston. Definitions in paediatric palliative care. Health Trends 25 (4):148-150, 1993.	'Consideration of care provision for children with life-threatening and life-limiting illness may be hampered by the lack of an agreed set of working definitions for such basic terms as: child, life-threatening conditions, life-limiting conditions, palliative care, respite care and hospice care. Describes how children with neurogenenerative, metabolic and genetically determined diseases have different palliative care needs to adults. Argues that it is rarely possible to offer an accurate prognosis. Offers definitions for the terms listed see paper.
Tadmor, C	C. S. Tadmor, S. Postovsky, R. Elhasid, A. Ben Barak, and M. B. Arush. Policies designed to enhance the quality of life of children with cancer at the end-of-life. Pediatr Hematol.Oncol. 20 (1):43-54, 2003.	A paper that talks about evaluating preventative intervention to enhance quality of life for children with cancer at the end of their lives. A description of policy and practice more then a study. Some interesting points about communicating with dying children and how they focus on life to the last weeks or days and are only ready to talk close to the end of their lives. Suggests that there are important differences between terminally ill and dying children
Walker, M	M. E. Walker and J. Jones. When children die: death in current children's literature and its use in a library. Bull.Med.Libr.Assoc 74 (1):16-18, 1986.	Paper describing the realistic and sympathetic descriptions of death in children's literature and how such books might be used to help children who are critically ill or bereaved. A list of useful books.

First Author	Reference	Summary of main points
While, A	A. E. While. The needs of dying children and their families. Health Visit. 62 (6):176-178, 1989.	Review of the literature in 1989. Presented as diagnosis and its aftermath, the illness, the final illness and bereavement. Implication of diagnosis for parents said to imply 'loss of a normal, satisfying childhood full of joyful moments and its replacement by a limited childhood characterised by hopelessness and pain.' Guilt of inherited disorders. Mismanagement of giving diagnosis 1. To one parent alone and 2 the general lack of information. Suggests the debate continues about whether the child should be told but more literature to indicate they should be. Child's understanding taken from Bluebond Langer. Brief mention of parents anticipatory mourning. Some literature around parents taking each day as it comes, enjoying time with the child. Highlights that health care provision is based on the assumption that families will adjust well without professional help to bereavement although literature indicates many parents would be better supported with bereavement counselling.
Wolfe, J	J. Wolfe, S. Friebert, and J. Hilden. Caring for children with advanced cancer integrating palliative care. Pediatr Clin North Am 49 (5):1043-1062, 2002.	Paper presenting a philosophy of communication with the child, family and all care providers involved. Argue for palliative care to be included along the whole disease trajectory - even if cure is possible. They include symptom control and communication in their concept of palliative care. They suggest that children with a terminal illness have a precocious understand of death and their own mortality. Provide an overview of the classical approach. They argue for timing in line with what the child initiates for themselves and what they call 'fleeting moments' that require 'immediate response' before they are lost but maybe with an indirect reply e.g. 'what makes you ask that?' but not with euphemisms. Many children are said to communicate best via non-verbal means e.g. artwork.
Woodgate, R	R. Woodgate. Adopting the qualitative paradigm to understanding children's perspectives of illness: barrier or facilitator? J Pediatr Nurs 16 (3):149-161, 2001.	Paper about the use of qualitative research methods for exploring children's perspectives and experiences of being ill. Refers to the studies of Bluebond Langer and theories of child development.

OUTLINE OF ETHICS PROCESS FOR REDESIGN AND REVIEW OF THE RESEARCH

In June 2002, the proposal was submitted to The South West Surrey Local Research Ethics Committee for consideration. Initially the committee were unable to approve the study but raised a series of comments and questions to help begin the process of review and redesign. Central to the committees concerns were the following issues:

i) The process of sampling and recruitment (see 'Sampling strategy' and 'Recruiting families').

ii) Care of families caring for dying children. It was made clear that families caring for a child who seemed to be very near the end of their life and who may only have had days or weeks to live were not going to be approached about participation in this research. In addition because the children's conditions could deteriorate at any time during the course of the study, consent to participate, was envisaged as on-going process of review between the researcher, child, their family and their senior care staff at the host children's hospice. The purpose of this collaborative strategy was to respond to each case on an individual basis so that families who agreed to participate could be certain that withdrawal from the study would have no impact on the care and support their child and family received from the host children's hospice.

iii) Anonymity for participants. Information gathered during this study was recorded and reported without using the names of the children or families who took part. However participants were informed that if in the course of the study any concerns arose about the child's safety or welfare these would be dealt with in the same way that they would be for any child cared for by the host children's hospice under the guidelines of the 'Child Protection Policy and Procedure'.

The child's medical diagnosis is not given because in some cases the rarity of diagnosis could lead to identification, therefore reference is only made to the child's prognostic category. Actual families are not presented because there were too many identifiable characteristics even with details omitted or changed. In addition the case is conceptual and it is not necessary to expose individuals in reporting the results of the study (see Data analysis). Grounded theory was used to analyse the mass of data collected. Only quotes and images relevant to illustrating the theory are presented and do not include photographs of individuals.

iv) The risk of emotional upset for participants and arrangements for dealing with emotional upset if it did arise as a problem (See appended 'Parents/main carers information sheet').

v) Inclusion of whole families rather than focus on one group for example, 'mothers'. It was explained that the aim of this research was to understand the experiences of the child as an individual, within the framework of their families, through understanding the range of persons and their perceptions as individuals and as a family. It was hoped that it would be helpful for family members to participate together, knowing that all their experiences were being valued individually as well as collectively.

vi) The potential impact on siblings or participating in the study. It was made clear that the objective was to ask children and siblings about their day-to-day experiences but not to bring them any agenda through that process. Consent to approach children and siblings about the research would be through parents initially followed by plans to work with children and their brothers and sisters, so that they could still decide not to participate or to withdraw from the study at any stage (see 'Consent as an on-going process').

vii) Creation of information sheets suitable for the age/ability range of children to be approached about participating in the study. In response to the Committees recommendation, information sheets for the children, young people and siblings of different ages were redrafted with advice from the Play Therapist at the host children's hospice. Separate information sheets were created for; Children, Young people, Young brothers and sisters, and Teenage brothers and sisters (these are appended to this report). These were to be available for the participants and used to supplement verbal explanations and discussions with myself. Overall the plan was to use them as a guide for talking, especially to very young children or those who did not read.

viii) The Committee requested 'topic guides' for interviews and they were created for interviews with; parents/main carers, children and young people, siblings, and people identified as the best sources of support (these are appended to this report).

In December 2002 I attended a committee meeting of The South West Surrey Local Research Ethics Committee with the Director of Care from the host children's hospice and my academic supervisor. This facilitated further discussion in person and was very constructive from all points of view. Ethics approval was given for this research at the committees next meeting.

Name	Relationship to the child	Age	Description of the child and family	Helping with	Child's opinions/my observations	Lessons
Fred	Child	12	Fred enjoys swimming and using the computer. He is looking forward to starting secondary school and lives with his mother, step father, brother, step brother and step sister. The family live in an adapted council house with a lift for Fred to get up-stairs. When Fred was only a few months old he was diagnosed with a relatively common but severe degenerative neuromuscular disorder that limits life expectancy to the late teenage years.	Review of books and my ideas for artwork – one session at his home.	Fred decided that 'A life Like Mine' - would be 'good' for his age group and 'little kids would enjoy' – 'All Kinds of Bodies' but 'I'm Special' - would 'help kids with disabilities to talk about themselves'. Fred's reaction to 'I'm Special' – was to look very intently at some of the issues, especially; 'People sometimes talk to mum as if I'm not there and I feel cross'. By coincidence his mother had left the room briefly. I asked Fred whether he found that sometimes people do talk to his mum instead of him and he nodded vigorously at me. His mother returned and he turned the page and we went on looking. Of the artwork ideas Fred said collecting things to illustrate a typical day and design a web page about you were the best ideas.	Fred was softly spoken, communicating lots non-verbally. **Fred's mother requested an indirect role for her son and I constructed an indirect role without seeking Fred's opinions.** Some images in the books encouraged Fred to tell me things that felt unrelated to the research initially, e.g. he was interested in an image of a caravan and it emerged that he missed a family holiday because of poor wheelchair access and the family hoped to buy a caravan to address this problem. I learned no to judge his starting place too quickly.
Jennifer	Sister	6		Review of books	Jennifer really enjoyed 'All kinds of bodies' reading it with her brother, mother and me.	Although Jennifer enjoyed the book it did not lead her to talk about herself or her brother in any detail.
Penny	Mother			Review of information sheets and topic guides	Penny looked at the books and artwork ideas with us. She had read the information sheets and topic guides and had no comments or suggestions. Penny began to share her own story of Fred's birth and diagnosis.	Parents have a complex and vital role in this research. Their consent and active participation may facilitate and regulate the role of child participants. Space for the parents own story will be crucial.

Table 1: Analysis of pilot data: Themes related to conceptual framework and implications for data collection procedures

Name	Relationship to the child	Age	Description of the child and family	Helping with	Child’s opinions/my observations	Lessons
Pete	Child	4	Pete enjoys playing with his siblings; he lives with his parents, two stepbrothers, and twin sister. Pete’s parents act to raise awareness of the needs of the children with special. The family live in a small four-bedroom London council house. Pete was diagnosed at birth with a rare condition that affects multiple systems of the body. Pete had severe life-threatening disease.	Met with the researcher but not actively participating in the pilot study	N/A	Pete’s parents were keen for the family to take part in the main study and so I did no more then meet the younger children. Pete’s mother suggested that I should see each child alone so they would get opportunities to talk to me on a one-to-one basis
Leah	Sister	4		Met with the researcher but not actively participating in the pilot study	N/A	See above
Philip	Brother	11		Given an explanation of the study and asked for his opinion.	Expressed interest in some of the methods especially photography.	Philip and Gary were enthusiastic about individual choice of artwork ideas for siblings. Philip had less to say but indicated his interest with eye contact and smiles.
Gary	Brother	10		Given an explanation of the study and asked for his opinion.	Especially interested in drawing and talking, ‘like being a journalist, cool!’	Gary clearly understood the aims of the research and sought clarification of his own place ‘oh about living with Pete for me but maybe not about me wanting to be an astronomer?’
	Mother			Review of research objectives.	Decided to participate in the main study	Parents have a complex and vital role in this research. Their consent and active participation are central. Space for the parents own story will be crucial
	Father			Review of research objectives.	See above	See above

Table 1: Analysis of pilot data: Themes related to conceptual framework and implications for data collection procedures

Name	Relationship to the child	Age	Description of the child and family	Helping with	Child's opinions/my observations	Lessons
Carol	Child	16	Carol lives with her parents in a residential area of Surrey. She enjoys music, videos and going out for a drive with her family. Carol attends residential school where her father says she is learning new skills and gaining some independence. Carol's brother attended boarding school form the age of 13 and is now at university. Carol's parents both studied law and work fulltime. Carol was diagnosed with an extremely rare complex seizure disorder in the first month of life.	Three sessions of participant observation; two at the hospice and one at home.	At the hospice Carol interacted with and responded to certain members of staff more then others and she was a little possessive of those she liked best. Carol did make choices that shaped her time at the hospice e.g. she wanted to watch a familiar video and chose who to sit next to for lunch [calling them with her finger, placing her hand on the seat beside her and pushing away unwanted 'others']. However I was not clear about the boundaries of her abilities to choose or make requests, e.g. it was a sunny day but she did not ask to go outside. I asked Carol 'do you like coming here?' and she looked at me quizzically saying 'yes?' so I asked 'what do you like best?' and she replied 'daddy, home!' grinning at me.	Participant observation, provided opportunities to witness Carol's experiences of visiting the hospice and in doing this I began to glimpse her personality. The time I spent alone with her gave me some basic observations to share with her father who was able to offer me a more detailed and complex picture. Carol was able to tell me that whilst she enjoys visits to the hospice, her family and home are important to her.
Steven	Father			Interview and review of research objectives especially for children who communicate non-verbally	Steven took part in a detailed interview about Carol and his experiences as her father. He listened to my basic observations and complimented them with his own insights. For example: He confirmed that Carol can communicate basic needs and wants but is also dependent on other people suggesting activities to increase her range of choices.	Again Steven underlined the place of parents in the research design. Firstly to facilitate their child's/children's participation and secondly to share their own story.

Table 1: Analysis of pilot data: Themes related to conceptual framework and implications for data collection procedures

Name	Relationship to the child	Age	Description of the child and family	Helping with	Child's opinions/my observations	Lessons
Primzanna	Child	3	Primzanna enjoys opportunities to stretch out and move her legs about. She is tall and heavy for her age. She goes to school and lives in a small council house with her parents, three sisters and older brother. The family came to England as refugees, to escape the troubles of their own country and seeking medical help for Primzanna, who suffers from a neurologically degenerative condition of unknown origin with epilepsy.	Short period of participant observation at home and at the hospice	I met Primzanna very briefly at the hospice, she was on her way out with staff and other children to the local shopping centre. When I visited the family home she was in bed resting.	It is not easy to tell how much Primzanna is aware of because her responses are very limited. Vocally she makes what appear to be random sounds. She makes eye contact for only a few seconds at a time and does not appear to use facial expressions to communicate. I felt that participation for a child with such limited communication abilities would be limited to the role and participation of family members, perhaps especially siblings.
Fulsanna	Sister	14		Photography and writing, making a poster and interview	Fulsanna took photographs and later construction of a poster about 'visiting the hospice'. During the interview; she sat as far away from the tape as possible and encouraging her sisters, 'you say'.	I was struck by the reticence of all three sisters. I felt I needed a broader selection of open questions for them. Unlike the adult participants across families these children were not waiting to share their story, they even looked surprised that I should ask.
Matina	Sister	12		Drawing and writing, making a poster and interview	Matina was willing to draw and make a poster but she opened up most when we were alone together, "The thing is Primzanna just needs our mum that's all she needs."	See above
Prea	Sister	7		Drawing, poetry, making a poster and interview	Prea began to comment on the 'lack or care at home' but couldn't find the words – Fulzanna helped her 'They just come here and look at Primzanna … they don't help my mum!'	Prea prompted her oldest sister to share a criticism of support services and this made it useful to see the sisters together. In other respects I felt each of the girls would have told me more on a one-to-one basis.
Martha	Mother			Review of the research objectives	I told Martha that the children didn't have very much to say and she commented; "Maybe they are not used to being asked but it is important to try and let them say?"	Martha shared her own story and endorsed the objective to hear from the children at the heart of the research.

Table 2: Analysis of pilot data: Themes related to conceptual framework and implications for data collection procedures

The Being Me Project: An exploration of the experience of being or caring for a life-limited child.

Topic Guide for Working with Parents or Main Carers

These headings are a general guide to the subjects that the parents will be asked to help us understand about 'Being me and having a child called ...'.

Being me and having a child called ...

- This is me
- My family
  About my child called ...
  About my other children ...
  About my partner ...
- What makes me happy?
- The best help for my child ...
  The best help for me ...
  The best help for our whole family ...
  How would I change things to make the help better ...
- What makes me sad?
- What makes me angry?
- When my child is well
- When my child is ill
- What I can do
- What I'm unable to do
- Things I like to do with the family
- Things I like to do on my own
- You didn't ask me this but ...

The hope is that the parents will lead discussions about their day or their week. Like the children they may choose to creating/save materials that will help them describe their day or week to the researcher. As a general guide for them before they start the topics listed below will be discussed as ideas.

My day/my week

- When I woke up ...
- The morning
- Things I did
- Lunchtime
- The afternoon
- The evening
- The night

- I went to 'X' with my child.
  Did I want to go?
  Did I feel it would help?
  How did it go?
  It was helpful that ...
  It was not helpful that ...
  The worst thing was ...
  The best thing was ...

- The 3 worst things about today/this week
- I was bored because ...
- The 3 best things about today/this week
- I laughed when ...

The Being Me Project: An exploration of the experience of being or caring for a life-limited child.

Topic Guide for Working with the Children and Young People

These headings are a general guide to the subjects that the children will be asked to help us understand about 'Being me' through discussion of their artwork. The researcher will communicate with each child/young person on an individual basis, based on their age and abilities using the themes listed as a general guide. If the child has particular needs related to communication the researcher will see them with someone they already know and can communicate with, in most cases this will be a parent or another person identified by the parents.

Being me

- This is me
- My family
- I am happy when ...
- It makes me sad that ...
- I get angry if ...
- When I'm well
- When I'm ill
- What I can do
- What I'm unable to do
- Things I like to do
- You didn't ask me this but ...

The hope is that the children will lead discussions about their day or their week by picking a particular day or week and saving materials that will help them describe their day or week to the researcher. As a general guide for them before they start the topics listed below will be discussed as ideas.

My day/my week

- When I woke up ...
- The morning
- Places I went to
- Lunchtime
- The afternoon
- Things I did
- The evening
- The night
- The 3 worst things about today/this week
- The 3 best things about today/this week
- I was bored because ...
- I laughed when ...

The Being Me Project: An exploration of the experience of being or caring for a life-limited child.

Topic Guide for Working with the Brothers & Sisters

These headings are a general guide to the subjects that the siblings will be asked to help us understand about 'Being me and having a brother or sister called ...' through discussion of their artwork. The researcher will communicate with each child on an individual basis, based on their age and abilities to discuss these general themes.

Being me and having a brother or sister called ...

- This is me
- My family
- About my brother/sister called ...
- What makes me happy?
- My brother/sister makes me happy because ...
- What makes me sad?
- My brother/sister makes me sad if ...
- What makes me angry?
- My brother/sister makes me angry when ...
- When my brother/sister is well
- When my brother/sister is ill
- What I can do
- What I'm unable to do
- Things I like to do
- You didn't ask me this but ...

The hope is that the children will lead discussions about their day or their week by picking a particular day or week and creating/saving materials that will help them describe their day or week to the researcher. As a general guide for them before they start the topics listed below will be discussed as ideas.

My day/my week

- When I woke up ...
- The morning
- Things I did
- Lunchtime
- The afternoon
- Places I went to
- The evening
- The night
- The 3 worst things about today/this week
- I was bored because ...
- The 3 best things about today/this week
- I laughed when ...

The 'Being Me' Project: An exploration of the experience of being or caring for a life-limited child.

Guide for participant observation

The purpose of participant observation in this study is for the researcher to understand more about the experiences of the participants, by being present for periods of time with the child and their family during episodes of their daily lives. There will no pre-established hypothesis to be tested. The researcher will be there to learn about the child and family's experiences and to get an overall feel of the situation through observation of the: -

- Setting
- Environment
- Details about events and what is happening during the period of observation
- Duration of events
- How does the child express him/herself?
- Details about interactions with the child
- What was said?
- Non-verbal communication
- How does the child respond to/communicate with adults that they know, peers, family members, strangers?

'THE BEING ME PROJECT'

INFORMATION SHEET FOR PARENTS & MAIN CARERS

1. Study title

The Being Me Project: An exploration of the experience of being or caring for a life-limited child.

2. Invitation paragraph

Your family are invited to take part in a research project. Before you decide it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information.

3. What is the purpose of the study?

The aim is to find out more about the day-to-day experiences of children with life-limiting conditions and their families. There is very little information from children and families themselves. Information about what it is like to live with this situation could guide professionals and support agencies about the sort of help that might suit families in the future.

4. Why has my child and our family been chosen?

We are asking a few children and their families if they would like to help us. The children will be different ages and suffer from different medical conditions.

5. Do I have to take part?

You do not have to take part. If you decide to take part you can still withdraw at any time, without giving a reason. These information sheets are to help you understand the project and what families will be asked to do. If you decide to take part you will be asked to sign a consent form.

These decisions will not affect the care and support you and your family receives from CHASE.

6. *Do my children have to take part?*

We will ask for your written permission to approach your child/children about taking part as well. They can also refuse or withdraw without giving a reason.

7. What will happen to us if we take part?

The researcher will meet you with (*name of identified CHASE care team member*), to discuss the project, so you can ask any questions. The researcher is using flexible methods for collecting information. Work with your child/children will be guided by you and their personal preferences or needs. You may be in contact with the researcher 'on and off' for a few weeks, but the aim is to complete the work with each family in about 5 sessions.

Members of you family will each be asked about - 'Being Me' and 'My day' or 'My week'. As parents you will be asked to help the researcher understand more about ***'Being me and having a child called ...'*** and ***'My day'*** or ***'My week'***. You can do this just by talking to the researcher or you might like to gather photographs or create art work to help you - but only if you would enjoy that! E.g. Mum, dad or another family member might choose to collect a series of photographs to illustrate 'My Week' and discuss them with the researcher.

With your permission the researcher will also spend an hour or two with you and your child, you might decide this session should be for example in hospital, at your home, or another setting. The researcher is spending this time with you to try and understand more about the experiences of your child and family by being there and observing things as they happen.

With your agreement it would be helpful for the researcher to have access to any relevant documents like your CHASE family notes or other relevant documents e.g. if you have taken part in a newspaper interview this would be a relevant document to share.
You will also be asked to name two people who support your child and family best and they will be asked to take part in an interview.

8. What about my child or children, what would they be asked to do?

The researcher will not approach your children without your permission. With your permission she will be introduced to them by a CHASE care team member who knows your family well, to talk about the project and ask if they would like to join in. Before meeting your child/children the researcher will need to talk to you about how you already talk to your child about their condition and explain any issues that come up or changes in their health. In the same way the researcher will want to understand how you already explain things to your other children (if you have any). This information will help the researcher to talk to your children about their experiences.

Like you they will be asked about 'Being me' and 'My day' or 'My week'. The researcher has a one-page guide for working with the children you might want to talk to the researcher about the general themes that will be covered. The children who take part can choose to create artwork to help them talk to the researcher. There are simple information sheets for the children that you will be given copies of to read and think over.

9. What are the possible disadvantages of taking part?

Taking part in this study will have no impact on any care or treatment that your child may be receiving currently or could receive in the future from CHASE Children's Hospice Service. However as parents you may have some of the following questions or concerns.

- ***What if we feel up-set or distressed by taking part in this research?***

Taking part in this study could raise some difficult questions for you and or your child or make you feel up-set or distress. This may happen during the study or sometime in the future. We wish to prevent or minimise any distress, but if you or any family member should require any extra emotional support now or in the future please do not hesitate to contact your CHASE identified care team member at any stage, even if you have reduced your level of contact with CHASE. CHASE does have access to various types of emotional support and therapy services that could be put in place to help you.

- ***What sort of help and support can you offer to a child who takes part?***

Your family's identified care team member will continue to be your first point of contact and main support. They will be aware that your child and family are taking part in this study and the researcher will work with them to identify extra support for any family member or child who needs it.

- ***Will you be giving detailed information to my child/children about their condition?***

No. The purpose of the project is to discover more about the daily experiences and issues faced by children living with a life-limiting condition and what they choose to share. Inevitably this may raise questions for them about their medical condition and future. If a child raises questions like these they/the issues they raise will be referred you and their identified CHASE care team member, who will provide/identify appropriate support.

- ***My child does not use speech how can they take part in this study?***

Children taking part in this study may be very young, use non-verbal methods of communication or their level of comprehension may be difficult to assess. The aim is to include children whatever their needs, recognising them all as valuable people. Every child has the right to refuse to take part/withdraw from the study. If your child is unable to communicate this kind of decision and you would still like them to join in you will be asked to take on that role for them and let us know when you feel that they should withdraw from the study, you do not have to give any reason for your decision.

Every effort will be made to gather information from your child or from their point of view using their preferred method of communication, observation and involving people who know them well.

10. What are the possible benefits of taking part?

You and your family are unlikely to benefit directly from taking part in this study for example the research process does not include any counselling techniques. Information that you share is likely to be helpful for various care agencies who may gain better insight into your situation.

11. What happens when the research study stops?

When the process is over the researcher will meet you all one last time to mark the end of the study and thank you for your time and effort. In time you will have a chance to review the main report and the researcher will talk to you about this at a later date.

12. What if something goes wrong or if we have any compiaints?

If you wish to complain, or have any concerns about any aspect of the way you have been approached or treated during the course of this study, the normal CHASE Children's Hospice Service complaints system is available to you.

13. Will my taking part in this study be kept confidential?

The information gathered during this study will be recorded and reported without using the names of the children or families who take part. However if in the course of this study any concerns are raised about the child's safety or welfare these will be dealt with in the same way that they would be for any child cared for by CHASE Children's Hospice Service under the guidelines of the 'Child Protection Policy & Procedure'.

14. What will happen to the results of the research study?

This study is being completed as part of a PhD and the results will be described in a thesis. You will be given the opportunity to see this material and talk to the researcher about it. Although it will not name any children or families who have taken part, you may feel that you/your family could be identified from something described, which seems especially unique about you. If you have any such worries please discuss them with the researcher. She will work with you and express things differently so that you feel confident that you and your family can not be identified.

In the future articles about specific issues are likely to be published in medical, health and social care journals and maybe presented at conferences. All this information will be based on the above materials and presented anonymously and no child or family will be named in any of these publications. In future years we will endeavour to contact you to ask whether you would like the opportunity to see material before it is published.

If you have other ideas about how the results could be shared or would like to know more in the future or obtain copies of published materials, please get in-touch with CHASE Children's Hospice Service. Please note research like this takes a long time and results may not be published for a few years.

15. Who is organising and funding the research?

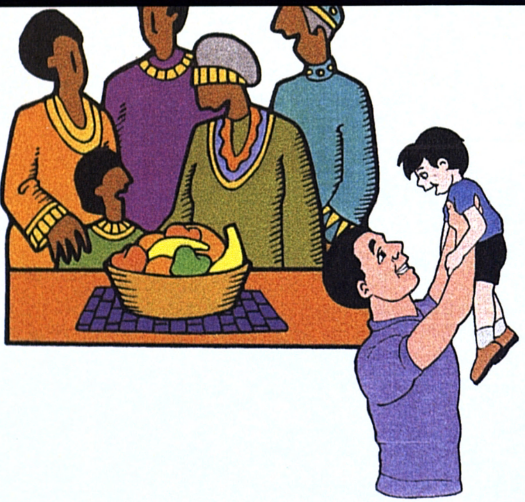
The researcher is an employee of CHASE Children's Hospice Service and has been awarded a Fellowship by The PPP Foundation to carry out this research. A senior tutor from Southampton University will supervise her.

16. Who has reviewed the study?

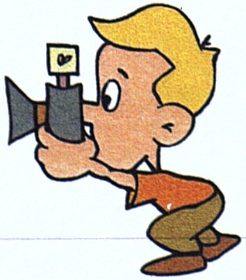
The South West Surrey Local Research Ethics Committee has reviewed this study.

17. Contact for further information

For further information please contact the researcher, Toni Menezes, Research Fellow, or your families named care team member at CHASE Children's Hospice Service.



The
Being Me
Project

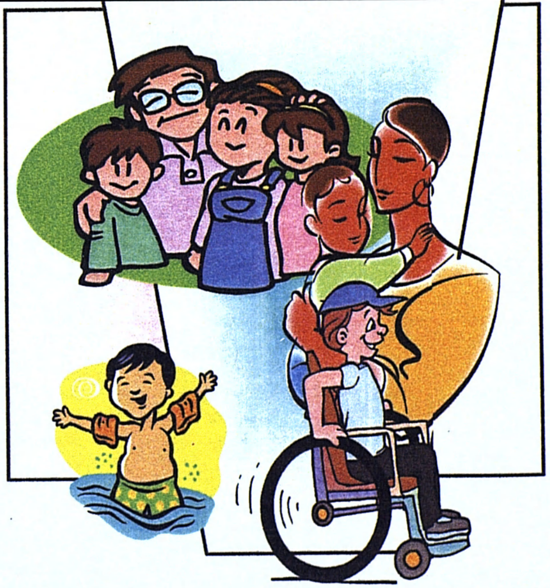


The
Being Me
Project

CHASE
hospice care for children

CHASE Children's Hospice Service

Www.chasecare.org.uk



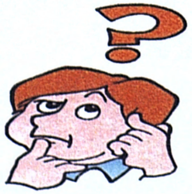
The
Being Me
Project

The Being Me Project

What is 'The Being Me Project'?

This project wants to find out more about what it's like for children and young people to live with a serious illness or disability that is difficult to treat or cure. You and your family could help by telling us more about this.

Why me?



We can't talk to everyone. So we are asking a few children and their families who use CHASE services, whether they would like to help us.

Do I have to take part?

You do not have to take part. If you say yes at first but then change your mind that's fine, just let us know. We only want children and young people to take part in the 'Being Me Project' if they really want to.

Who is doing this project?

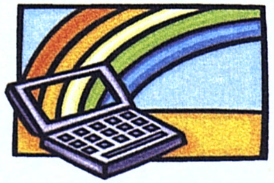
Toni Menezes who works at CHASE is doing the project. Her job is to work with the children on the project. Then to tell doctors and other people who look after children what we found out. She will be writing reports and giving talks for professionals.

First you would meet Toni with your CHASE Care Team Member to talk about the project and ask any questions.

Then you and your family will work with Toni.

You will be helping with 2 topics 'Being me' and 'My day'. It might be fun to make things, talk, draw pictures or take photos. Toni has some ideas and things to use. If you have more ideas that's great!

'Being me'



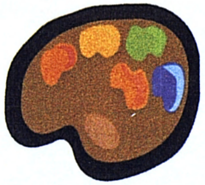
What about a poem, or a picture, or a model?

'My day'

Choose a day to tell Toni about. It might be helpful to use photos, or pictures and cuttings, or other things you have collected!

During the project Toni will spend a little time with you and your family, seeing how your day goes. Just a place you would normally be anyway. You and your family will get to choose the place.

You will be making things for the project so that other people get a chance to share them and enjoy them, so it will be important for Toni to keep the things you make. We will give you photos of your artwork for you to keep.



It's private and confidential!

Toni will be writing a report about this project, but she will not use your name or any of your family's names because what you tell us is private. But you can tell who ever you want.

What happens when the project stops?

When the project is over Toni will meet you and your family one last time to thank you for all your help.

Who can I talk to if I have any questions or worries about this project?

My Care Team Member

____

Telephone ______________

The researcher, Toni

Telephone ______________

'THE BEING ME PROJECT'

INFORMATION SHEET FOR CHILDREN

What is 'The Being Me Project'?
This project wants to find out more about what it's like for children and young people to live with a serious illness or disability that is difficult to treat or cure. You and your family could help by telling us more about this.

Why me?
We can't talk to everyone. So we are asking a few children and their families whether they would like to help us.

Do I have to take part?
You do not have to take part. If you say yes at first but then change your mind that's fine, just let us know. We only want children and young people to take part in the 'Being Me Project' if they really want to.

Who is doing this project?
Toni Menezes who works at Christopher's is doing the project. Her job is to work with the children on the project. Then to tell doctors and other people who look after children what we found out.

What would I have to do?
Firstly, Toni will meet you with (*name of identified CHASE care team member*). To talk about the project, so you can ask any questions.

After that you and Toni will be working together on 2 topics. 'Being me' and 'My day'. It might be fun to make things, talk, draw pictures or take photos. Toni has some ideas and things to use. If you have more ideas that's great! Sometimes the things we have made or collected help us to explain to other people, because they can look and ask questions as well.

'Being me' – What about a poem, or a picture, or a model!

'My day' – You will choose a day to tell Toni about. It might be helpful to use photos, or pictures and cuttings, or other things you have collected!

Once during the project Toni will spend a little time with you and your family, seeing how your day goes. Just a place you would normally be anyway. You and your family will get to choose the place.

What happens to the things I make?
You will be making things for the project so that other people get a chance to share them and enjoy them, so it will be important for Toni to keep the things you make. We will give you photos of your artwork for you to keep.

It's private and confidential!
Toni will be writing a report about this project, but she will not use your name or any of your family's names because what you tell us is private. But you can tell who ever you want.

What happens when the project stops?
When the project is over Toni will meet you and your family one last time to thank you for all your help.

Date: 14/11/2002 **Version:** 2

'THE BEING ME PROJECT'

INFORMATION SHEET FOR YOUNG PEOPLE

What is 'The Being Me Project'?
This project aims to find out more about what it's like for children and young people to live with a serious illness or serious disability that is difficult to treat or cure. You and your family could help by telling us more about your day-to-day experiences.

Why me?
We can't talk to everyone. So we are asking a few children, young people and their families whether they would like to participate in this project.

Do I have to take part?
You do not have to take part. If you say yes at first but then change your mind that's fine, just let us know.

Who is doing this project?
Toni Menezes who works at Christopher's is doing the project. Her job is to work with the children and young people to ascertain their views. Then to share the information with other professionals who try to help families like your own.

What would I have to do?
Firstly, Toni will meet you with (*name of identified CHASE care team member*). To discuss the project, so you can ask any questions.

After that you and Toni will be working together on 2 topics. 'Being me' and 'My day' or 'My week'. You could contribute to the project by, talking to Toni, drawing pictures or taking photos. Toni has some ideas and things to use but if you have your own ideas that's great! The artwork that you create might help other people to begin to understand what it's like to be you.

'Being me' – What about a poem, or a picture, or a model!

'My day' or 'My week' – You will choose a day or a week to tell Toni about. It might be helpful to use photos, or pictures and cuttings, or other things you have collected!

Once during the project Toni will spend a little time with you, seeing how your day goes. Just a place you would normally be anyway. You and your parents will choose the venue it could be hospital, school or somewhere else if you prefer.

What happens to the things I make?
You will be making things for the project so that other people get a chance to share them so it important that Toni keeps them to use in the future. We will give you photos of anything you produce as a memento of your participation.

It's private and confidential!
Toni will be writing a report about this project, but she will not use your name or any of your family's names because what you tell us is private. But you can tell who ever you want.

What happens when the project stops?
When the project is over Toni will meet you and your family one last time to thank you for all your help and to talk to you about how we use the artwork and information from this project in the future.

Date: 14/11/2002 **Version:** 2

'THE BEING ME PROJECT'

INFORMATION SHEET FOR YOUNG BROTHERS AND SISTERS

What is 'The Being Me Project'?
This project wants to find out what it's like to have a brother or sister with a serious illness or disability that is difficult to treat or cure. You and your family could help by telling us more about this.

Why me?
We can't talk to everyone. So we are asking a few brothers and sisters whether they would like to help us with their families.

Do I have to take part?
You do not have to take part. If you say yes at first but then change your mind that's fine, just let us know. We only want children and young people to take part in the 'Being Me Project' if they really want to.

Who is doing this project?
Toni Menezes who works at Christopher's is doing the project. Her job is to work with the children on the project. Then to tell doctors and other people who look after children what we found out.

What would I have to do?
Firstly, Toni will meet you with (*name of identified CHASE care team member*). To discuss the project, so you can ask any questions.

After that you and Toni will be working together on 2 topics. 'Being me and having a brother/sister called ....' and 'My day'. It might be fun to make things, talk, draw pictures or take photos. Toni has some ideas and things to use. If you have more ideas that's great! Sometimes the things we have made or collected help us to explain to other people, because they can look and ask questions as well.

'Being me and having a brother/sister called .....' – What about pictures and cuttings, or photos, or a poem!

'My day' – You will choose a day to tell Toni about. It might be helpful to use, your drawings, or a model!

What happens to the things I make?
You will be making things for the project so that other people get a chance to share them and enjoy them, so it will be important for Toni to keep the things that you make. We will give you photos of your artwork for you to keep.

It's private and confidential!
Toni will be writing a report about this project, but she will not use your name or any of your family's names because what you tell us is private. But you can tell who ever you want.

What happens when the project stops?
When the project is over Toni will meet you and your family one last time to thank you for all your help.

Date: 14/11/2002 **Version:** 2

‘THE BEING ME PROJECT’

INFORMATION SHEET FOR TEENAGE BROTHERS AND SISTERS

What is 'The Being Me Project'?
This project wants to find out what it’s like to have a brother or sister with a serious illness or serious disability that is difficult to treat or cure. You and your family could help by telling us more about your day-to-day experiences.

Why me?
We can't talk to everyone. So we are asking a few brothers and sisters whether they would like to help us with their families.

Do I have to take part?
You do not have to take part. If you say yes at first but then change your mind that's fine, just let us know.

Who is doing this project?
Toni Menezes who works at Christopher's is doing the project. Her job is to work with the children and young people to ascertain their views. Then to share the information with other professionals who try to help families like your own.

What would I have to do?
Firstly, Toni will meet you with (*name of identified CHASE care team member*). To discuss the project, so you can ask any questions.

After that you and Toni will be working together on 2 topics. ‘Being me and having a brother/sister called ….’ and ‘My day’. It might be fun to make things, talk, draw pictures or take photos. Toni has some ideas and things to use. If you have more ideas that's great! The artwork that you create might help other people to begin to understand what it's like to be you.

‘Being me and having a brother/sister called …..’ – What about pictures and cuttings, or photos, or a poem!

‘My day’ – You will choose a day to tell Toni about. It might be helpful to use, your drawings, or a model!

What happens to the things I make?
You will be making things for the project so that other people get a chance to share them and enjoy them so it will be important for Toni to keep the things you make. We will give you photos of anything you produce as a memento of your participation.

It's private and confidential!
Toni will be writing a report about this project, but she will not use your name or any of your family's names because what you tell us is private. But you can tell who ever you want.

What happens when the project stops?
When the project is over Toni will meet you and your family one last time to thank you for all your help and to talk to you about how we use the artwork and information from this project in the future.

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My house; fun time sticker book (2004) Brown Watson. England
A book of illustration depicting the people and places in a child's home, the book has no text.

At school fun time sticker book (2004) Brown Watson. England
A book of illustration depicting the people and places in a child's school, the book has no text.

A life like mine: How children live around the world. A UNICEF book published by Dorling Kindersley (2002).
This book has approx. 130 large pages that show with words and photographs how children are leading their lives in different ways around the world. In 4 sections covering survival, development, protection and participation it covers a wide range of topics and issues related to the lives of children.

Of specific interest to this research it includes sections on: 'Every child deserves a healthy life' p36; 'Natalie (how a young girl is affected by asthma) p42; 'No child should be held back by a disability' p90; 'what is life like with a disability' p92; and 'Living with a disability' p94.

All kinds of bodies: A lift-the-flap book Emma Brownjohn (2002) by Tanga Books. London
This little book has lift-the-flap pages that show how people have different shaped hands, feet, faces and bodies. How some people wear glasses and others don't, how some people use crutches or wheelchairs to get around and others don't. It claims to show how we are all special, although different to each other.

I'm special Jen Green and Mike Gordon (2003) by Wayland Publishers. London
This book provides an introduction to the subject of disability for young children. It describes the feelings of disabled people in their everyday life and in their encounters with people around them.

Going to the hospital Fred Rogers (1988) A Paperstar Book. Penguin Putnam Books for Young Readers. New York
The book contains pages of coloured photographs showing children experiencing hospital for the first time. The text is simple and talks to children about hospital visits.

Going to the hospital Anne Civardi (2002) Usborne First Experiences. London
This is the story of a young boy's first visit to the hospital. Each page is full of detailed illustrations that show people, events and places in hospital.

Tomorrow I will feel better Ann De Bode and Rien Broere. Evans Brothers Limited. London
This is the story of a young girl who has heart surgery. The book has illustration and text about a complex experience.