

NAVIGATING UNCHARTED TERRITORY: EXPERIENCES OF FAMILIES WHEN A
CHILD HAS A NEURODEGENERATIVE LIFE THREATENING ILLNESS

by

ROSE GUILLER STEELE

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M.Sc., University of Toronto, 1994

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School
~~Department~~ of Nursing

The University of British Columbia
Vancouver, Canada

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Abstract

Children with neurodegenerative, life-threatening illnesses account for a significant proportion of children requiring palliative care. Most of their care is provided at home by their families. Yet, there is a paucity of research that examines families' experiences when a child is dying at home. This grounded theory study generated a contextually-grounded description of families' experiences of living with a child who has a neurodegenerative, life-threatening illness, and how those experiences changed over time; the impact on families; and families' perceptions of factors that influenced their ability to care for their children. Data were collected from eight families through observations and audiotaped interviews.

Families moved through a process of *navigating uncharted territory* as they lived with their dying child. This process occurred within the broader context of the *acute, curative health care system* and the *sociocultural environment*. The process was initiated by a precipitating event, such as one parent recognizing a deterioration in the child's motor skills. The emotions of fear, uncertainty, and grief gave impetus to the process. These emotions were very strong in the beginning. They gradually declined over time, but increased with subsequent precipitating events. Families lived much of their lives on plateaus of relative stability where they often felt alone and isolated from health care professionals. Inevitably, periods of instability originated in subsequent precipitating events in the process that led to families *dropping off the plateau*. Living with a dying child had a profound physical, emotional, and financial impact on families.

The process of *navigating uncharted territory* was characterized by four dimensions - *entering unfamiliar territory, shifting priorities, creating meaning, and holding the fort*. Parents used strategies within these dimensions to manage the physical, cognitive, and emotional work arising from the situation. These dimensions and strategies were constrained and facilitated by

four intervening conditions - *relationships with health care providers, availability of information, gender differences, and communication between parents.*

The theory generated in this study contributes to an understanding of families' experiences as they live with a dying child. This theory provides a framework from which appropriate, family-centred interventions can be developed to facilitate optimal palliative care.

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Chapter I: Problem

Background to the Problem

Over the past 30 years, the field of palliative care has become fairly well established with adults. Pediatric palliative care, however, is relatively new. It is only in the last 10-15 years that pediatric palliative care has received clinical and, even more recently, research attention. The basic principles of the palliative care movement are similar for both the adult and the pediatric populations: (a) to provide physical, emotional and spiritual care to the sick person and the family, (b) to promote the best quality of life, and (c) to provide bereavement care. As suggested by the leading pioneer in pediatric palliative care, Mother Frances Dominica from England, the role of pediatric palliative care is to offer friendship, support, and practical help to families (Dominica, 1987). In addition, complex symptom management is an integral part of pediatric palliative care programs (Goldman, 1998; McGrath, 1998).

Ideally, palliative care should be introduced early in the child's progressive, life-threatening illness (PLTI), preferably at the time of diagnosis (Frager, 1996). This early link with palliative care services allows an opportunity to promote palliative care principles, facilitates family-centred care that is responsive to the changing needs of families, and may help prevent crisis-oriented management. As the family's needs dictate, attention should shift from acute to palliative care that includes respite and terminal care, and then to bereavement care. Thus, health professionals not only provide bereavement care, but often work with grieving families for a considerable period of time before the child dies.

In adult palliative care, most patients have cancer and are expected to die within six months of admission to palliative care. Conversely, children in pediatric palliative care suffer from a wide variety of diseases and syndromes. About 20% of the children may have cancer, but many have progressive neuromuscular or neurodegenerative conditions, such as Battens disease or one of the leukodystrophies, that will eventually cause their death, though not necessarily in

the next six months (Ashby, Kosky, Laver, & Sims, 1991; B. Davies & Howell, 1998; Goldman, 1996). Consequently, the time between diagnosis and death is variable in these children. Many children live for a prolonged period of time, though they will not reach adulthood. Families often require respite care during this protracted period of time. The pioneers in pediatric palliative care (Dominica, 1987; Hunt, 1986) emphasized the importance of respite care for these families.

While there has been an increasing focus on complex symptom management in the past few years (Goldman, 1998; Liben, 1996; McGrath, 1998), little has changed in the commitment to respite care as an integral component of pediatric palliative care (B. Davies & Howell; Goldman, 1998). Respite care is acknowledged as essential for families (B. Davies & Howell; Goldman, 1996). It provides them with some relief from the unremitting, 24-hour care of their ill children. Respite care is usually provided for a relatively short period of time on an intermittent basis to allow families to have a break.

The numbers of children with a prolonged terminal illness are low when compared with those in the adult population. For example, there were 346 children who died from terminal illnesses in British Columbia over the three year period 1988-1990. At any one time, there are over 200 children in the province who are living with progressive, life-threatening illnesses (B. Davies, 1992). One estimate from the United Kingdom indicates that 1:1000 children may be affected by life-threatening illnesses (Goldman, 1996). Although the current numbers are relatively small, these children may pose substantial management problems and may use a disproportionate amount of in-patient health resources (Caring Institute of the Foundation for Hospice and Home Care, 1987). Additionally, the numbers of children with a PLTI are projected to increase as the incidence of life-threatening diseases rises and as advances in technology and medicine reduce mortality rates for conditions from which children previously died (Broome, 1998; B. Davies & Howell, 1998).

Home care, rather than in-patient care, is strongly supported for pediatric palliative care (Lauer, Mulhern, Hoffmann, & Camitta, 1986; Stevens, 1998). Most of the care for these children is typically provided at home by their families over an extended period of time that may last years rather than months (Burne, Dominica, & Baum, 1984; Goldman, 1998; Stevens). The home provides a secure, familiar, and comfortable environment for the child (B. Davies & Howell, 1998). It allows parents to feel more in control and is less disruptive to family life (Martinson, 1993). However, the impact of caring for a child with a PLTI can be substantial. Care is often provided to the detriment of the individual caregivers and to the family itself (Folden & Coffman, 1993; Stevens).

Children exist within a family, so the child's illness affects the whole family. In turn, the family's response to the illness has an important impact on the ill child (Friedman, 1998; Leonard, Enzle, McTavish, Cumming, & Cumming, 1995). Therefore, experts agree that care of these children must also include their families. The ill child, parents, and siblings must be seen as the central focus of care. They must also be involved in all aspects of care and decision making (Martinson, 1993). Parents want and expect to take care of their child. They may want to be in control even more so than families of terminally ill adults (B. Davies, 1992). Families of dying children tend to be large because parents and grandparents are still alive, siblings live at home, and friends are part of the larger affected group. So meeting the needs of these families often requires more time, skill, and expertise than is needed for families of older patients.

Family stress is at its highest when a member is entering or leaving the family system and homeostasis or equilibrium is disturbed (Friedman, 1998). Thus, families with a dying child are at risk for suffering the emotional and physical effects of stress. The increased media attention on such families in recent years has documented some of the stresses families face, as well as the consequences. For example, a mother in Canada was charged with starving her ill child to death, and in another family, the father was convicted of manslaughter in the death of his severely

disabled child. The problems faced by these families have been thrust into the public eye and have led to increasing debate over how professionals can best support families. Although no morbidity statistics are available, caring for a child with a PLTI at home has been associated with emotional, psychological, and financial distress in parents (Davies, 1996; Diehl, Moffitt, & Wade, 1991; Stein & Woolley, 1990). Parents may be anxious, suffer from insomnia, and be socially dysfunctional; some may start to use or increase their use of tranquillisers, anti-depressants, or cigarettes to relieve their anxiety (Stein & Woolley). Parents also struggle with trying to balance the demands of their ill child with the needs of healthy siblings. In addition, the ill children may experience emotional problems, such as anxiety and unhappiness. Siblings also experience distress when a brother or sister is dying. Indeed, in some families, siblings may have more problems than the dying child (Stein & Woolley). Emotional and behavioural problems, such as difficulty in dealing with their school work or increased aggression or withdrawal, may occur.

Bakke and Pomietto (1986) noted that health professionals want to support grieving families. Yet, they are often unaware of the specific elements of support, such as the type of information needed or the location of care, that would be most beneficial for families. Additionally, health professionals, such as nurses, often view themselves as advocates for patients and families. As such, professionals must be capable of presenting information and alternatives to families so families can make the choice that is most appropriate for them.

Anecdotal data suggest that although families often require substantial attention, information, and support, there are important differences between the perceptions of families and staff in how well these needs are met. For example, bereaved families often say, "If only they had told me to expect..." (that the child would hemorrhage, or what would occur at the time of death, and so forth). Yet, healthcare professionals will point out that those issues and information had been discussed repeatedly during the child's illness. When provided with clear evidence that

the issues were discussed and information given, families tend to say, "Oh, yes. I'd forgotten that." They cannot retrospectively identify what else the health care professionals could have done to have made the experience any easier. Little is known about the kinds of information that are useful, when families hear or do not hear information, what kinds of support are desirable, or how that support should be given. There may be times or stages when "teachable moments" occur in the lives of families, or families may give cues that could alert health care professionals to intervene in a particular way. No research is currently available to identify such events.

Family involvement in care of a child with a PLTI is complicated by the fact that death and dying are generally taboo subjects in the United States of America (USA) and Canada (Fulton, 1994). In these North American societies, people seldom think about death anymore, especially a child's death. North Americans are reticent to talk about the meaning of death in human life, about appropriate responses to death, or about the place of rituals and communal bonds in the face of death. There is no general culture for dealing with issues around death. Society hides death, such as with the almost exclusive use of funeral homes in North America (Walsh & McGoldrick, 1988). North Americans often seem to act as if death is just another disease to overcome. The logic that death is a constant enemy is at odds with the belief of many clinicians that death is part of living. Moreover, the USA has been described as unusual among developed countries in its attempts to try to conquer death (Callahan, 1995), though Canada also fits with this description. Some countries seemingly have less difficulty in caring for the dying - they do not struggle with the clinical, moral, and legal problems seen in the USA and, to a somewhat lesser extent, in Canada. In North American culture, physicians often do everything that is possible, but when they are unable to do more then they leave the patient to nurses (Callahan). Many professionals have a particularly difficult time in identifying a child as terminally ill. They refrain from moving from curative to palliative care at an appropriate time (B. Davies & Steele, 1996; O'Gorman, 1998). Although the palliative care movement has led to

many changes in the care of dying patients, too many health professionals remain afraid of death. Therefore, professionals cannot always provide the support families need because of their own insecurities and fears.

Over the 20th century, death has moved from the realm of the sacred to the secular, from the private to the public, and from the natural to the artificial (Callahan, 1995; O'Gorman, 1998). Medicine takes the view that death can be averted or treated, as can be seen in the rapid increase in technology and intensive care units. Whereas people used to die at home with their families, dying in hospitals and being cared for by professionals has become the norm. Since people are no longer confronted by the death of older family members at home, most young adults have gained little experience of death and dying. Therefore, young parents of children with life-threatening illnesses have not had to deal with many deaths before, and almost certainly not a pediatric death (B. Davies, 1992). This lack of experience with death and dying can exacerbate a situation that is already fraught with real and potential difficulties. In addition, families are usually developmentally and chronologically young, and they may need considerable attention, skill, and time to help them survive the experience (B. Davies).

Statement of the Problem

There is a lack of knowledge about the experiences of families where a child has a progressive, life-threatening illness. Unlike adult palliative care, a substantial percentage of children in pediatric palliative care die from neurodegenerative, life-threatening illnesses (NLTIs). Health professionals have barely begun to document the effects on families of caring for a child with an NLTi. Little is known about the most appropriate interventions for these families. Retrospective accounts of a child's illness have not always been accurate or even possible. Therefore, in addition to retrospective data collection, a prospective approach is important to capture family experiences as they evolve.

Purpose

The purpose of this study was to enhance understanding of the experiences of families with a child who has an NLTI. The knowledge generated by this study and its contribution to developing theory will be used to promote effective palliative care of these families. Specific aims of the study were to: (1) describe families' perceptions and experiences of living with a child who has a neurodegenerative, life-threatening illness, and how those experiences changed over time; (2) describe the impact on the family of living with a child who has a neurodegenerative, life-threatening illness; and (3) describe families' perceptions of the factors that influenced their ability to care for their child who has a neurodegenerative, life-threatening illness.

Summary

Increasing numbers of children with progressive, life-threatening illnesses are being cared for at home by their families. Health professionals can provide support to families, but they do not have a clear understanding of what kind of support is appropriate or how to provide that support to these families. Living with a child who has a PLTI may have a profound impact on families. Many families may experience psychological, physical, financial, and social disruptions that may affect their quality of life. Health care professionals are in a position to provide interventions that will promote the health and welfare of families, if they know the type of interventions to offer. Although palliative care has been developing rapidly in the past decade, there is still a dearth of research in this field, particularly in the pediatric area. It is imperative that suitable interventions are uncovered and developed to provide guidance to practitioners. This research study focused on the experiences of families when a child has a PLTI, specifically a neurodegenerative illness. A relatively high percentage of children requiring palliative care suffer from neurodegenerative illnesses, yet very little research has been completed in this area.

The findings of this research provide a foundation for identifying interventions that will enable health care professionals to provide the most appropriate care to such families.

The remainder of this dissertation follows a fairly standardized format. The conceptual framework that underlies the study is introduced in Chapter II. This framework sets the context in which the research questions were asked. Related literature is reviewed in Chapter III to provide an understanding of what is currently known about families where a child has a progressive, life-threatening illness, especially a neurodegenerative disease. In Chapter IV, the research design, procedures, and implementation are explained. The findings of the study are presented in Chapter V. The experiences of families when a child has a neurodegenerative, life-threatening illness are described and conceptualized through the process of *navigating uncharted territory*. This newly developed theory is placed within the larger context of current research and literature in Chapter VI. Finally, the study is summarized and implications for practice, education, and research are offered in Chapter VII.

Chapter II: Conceptual Framework

A conceptual framework underlies a study problem and supports the rationale for conducting a study. It describes the perspective from which one views the world, and gives guidance for understanding the context in which one studies phenomena. A conceptual framework is not synonymous with a theoretical framework. In this study, grounded theory methodology was used to develop a contextually-grounded theory about the experiences of families when a child has a neurodegenerative, life-threatening illness (NLTI). Clearly, the use of a previously developed theoretical framework would have been inappropriate for this purpose. However, researchers do not begin a study without some understanding of the context within which the research questions will be asked. Four major concepts formed the context for this study. The first concept concerned children and life-threatening illnesses, the second concept was related to families, and the third concept was coping. The focus of this study was where these three concepts intersected within the broader context of grief. In this chapter, the researcher's perspective of these four major concepts will be described in order to delineate the context in which the study was conducted.

Perspective on Grief

Families with a child who has a progressive, life-threatening illness live with the threat of death and dying every day. Death means the loss of many things - the loss of this life, of self as a person, of tomorrow, of those we love, and of all we know. In families with a dying child, losses are felt by every member of the family. Not only must the parents prepare themselves for their child's death, but the ill child and siblings must also deal with this reality. Therefore, it is important to understand the relevant theories of death and dying that affect both families and those who provide care to them. In this study, grief was thought of as a process, rather than as stages. Families were not expected to move through pre-determined stages. Instead, the researcher sought understanding about the process of grief.

Conceptualizations of Grief

In early conceptions of grief, clinicians and theorists viewed grief as linear and time-bound (Miles & Demi, 1994). Terms such as grief, grieving, and mourning were used synonymously. Uncomplicated grief was seen as a progression through stages of shock and disbelief; developing awareness of loss; and prolonged recovery. The work of uncomplicated grief was expected to take 4-6 weeks to complete (Miles & Demi; Teel, 1991). If mourning was delayed, chronic, exaggerated, or masked, then the grief response was said to be abnormal. Consequently, the extended grieving typical of families of a dead child was seen as pathological and requiring psychiatric assistance or counselling.

Although there had been a number of theories proposed about grief, little attention was paid to death and dying by professionals and lay people until Elisabeth Kubler-Ross published *On Death and Dying* in 1969 (Kubler-Ross, 1969). This book was instrumental in bringing the staged model of grief into public and professional awareness. According to Kubler-Ross, people go through five different stages when they are faced with dying: denial and isolation; anger; bargaining; depression; and acceptance. Although she wrote that, "These means will last for different periods of time and will replace each other or exist at times side by side" (p. 122), many professionals and lay people viewed the stages as sequential. Consequently, patients were expected to move through these stages until they finally accepted their death. Many professionals and lay people continue to work today with this notion of linearity and time-bound grief. In recent years, however, the linear model has been questioned by some clinicians and theorists (Cowan & Murphy, 1985; Worthington, 1989; 1994). Questions have been raised particularly in relation to the length of time considered "normal" to continue grieving, and the assumption that failure to achieve resolution of grief is abnormal. The five-stage model attributed to Kubler-Ross is not supported by research and has been called superficial, inadequate, and misleading (Corr, 1993). There has been no demonstration of the existence of the stages, nor evidence that people

actually move from stage one through to stage five (Buckman, 1998). Instead, humans cope in many ways, not just these five. There are no prescriptive or obligatory ways in which people should cope with death and dying. There is no "right" or "wrong" way to die. Coping with dying is a richer and more complex process than merely progressing through stages.

Buckman (1998) proposed a three-stage model in which he argued that individuals facing death react in ways that are consistent with their character and their previous coping patterns. Reactions are, therefore, not a product of the diagnosis or the stage of the dying process. In addition, Buckman proposed that progress through the dying process is marked by resolution of the elements in emotions that are resolvable, rather than by a change in the type of emotions. His model incorporates three stages: initial (facing the threat); chronic (being ill); and final (acceptance of death). When emotions in the first stage are resolved, then an individual moves into the second stage. Not everyone reaches this second stage. Buckman suggests that those individuals require professional help. In the third stage, emotions are less intense and the individual accepts impending death. While this model is an improvement on Kubler-Ross' model (Kubler-Ross, 1969), it continues to present a mechanistic and unidimensional approach to a multidimensional process.

Many theorists and clinicians now conceptualize grief as a process, not as stages (Attig, 1991; 1996; Cowles, 1996; B. Davies & Eng, 1998; Gyulay, 1989). According to Attig (1996), grief is the specific emotion that one feels in reaction to loss. This emotion engenders helplessness and passivity because the emotion grief is essentially irrational. Normally, emotions are motivating. But grief cannot motivate because there are no recognizable means to the end desired - wishing will not prevent death or return the deceased person to life. Attig suggested that the depth and power of grief may derive from this lack of motivational force. Furthermore, in contrast to the passive, standard conceptions, Attig viewed the process of grieving as an active, complex process that presents challenges and opportunities. An active process may be more

accurate descriptively and may capture more of the complexity of bereavement than a more passive view. It may also be more powerful in promoting self-understanding and in providing better direction for those who would help the dying and the bereaved.

There is some empirical research to support a dynamic conceptualization of grief. Focus group participants from a variety of cultural backgrounds were asked to draw on their own personal experiences of grief. Their definitions of grief were similar to those in the professional literature (Cowles, 1996). Irrespective of their culture, they viewed grief as a dynamic, pervasive, and highly individualized process. Participants agreed that culture or cultural heritage is a key component of the context in which people respond to actual or potential health problems. They also believed that, though mourning rituals or traditions may be culturally defined and proscribed, each person experiences grief in his or her own way. In addition, grief occurs universally across all age groups. In contrast to the professional literature that often emphasizes temporal and behavioural limits to grief, the research participants did not impose limits. Neither were they judgmental about others' experiences of grief. Others' experiences were viewed as simply different, not as wrong. Most agreed that the progression of grief is unpredictable and that changes over time vary. In addition, grief does not necessarily have a particular end-point.

McClowry, B. Davies, May, Kulenkamp, and Martinson (1987) provided further support for this view of grief. They used grounded theory methods to study 49 families who were interviewed 7-9 years after a death following childhood cancer. Many parents and siblings were still experiencing pain and loss at the time of interview. Despite similarities in experiences related to the initial loss of the child (i.e., in that it had created an "empty space"), three patterns of grieving responses were noted: "getting over it;" "filling the emptiness;" and "keeping the connection." Families stated that time lessened the pain. In addition, the sense of emptiness changed over time. The "empty space," however, was likely to recur at specific times such as holidays and anniversary dates like the child's date of birth or death.

Chronic Sorrow

Recurring waves of grief or chronic sorrow have been observed in parents of children with mental or physical disabilities. This pervasive, recurrent sadness was first characterized by Olshansky (1962) as a normal response to disruptions in anticipated normalcy. Clinicians and researchers have reported that families' ongoing losses may also be associated with feelings such as sadness, anger, depression, confusion, fear, anxiety, embarrassment, and isolation that vary in intensity among different persons, across situations, and at different times within an individual (Fraley, 1990; Lindgren, Burke, Hainsworth, & Eakes, 1992; Phillips, 1991; Teel, 1991). Further research has expanded the concept of chronic sorrow to individuals experiencing a variety of loss situations, as well as to their family caregivers (Eakes, 1993; Eakes, Burke, Hainsworth, & Lindgren, 1993; Hainsworth, Eakes, & Burke, 1994; Lindgren, 1996). Chronic sorrow is based on the premise that people may experience recurrent episodes of grief following a loss. The cyclical nature of chronic sorrow means that each episode of grief wanes over time. Periods of happiness are interspersed between episodes of grief, so grief does not usually become incapacitating (Lindgren et al.). When a child has an NLTi, a family experiences losses over many years as the child's condition worsens. Their experiences of grief associated with these losses are not well understood. Moreover, parental feelings of recurrent grief may resurface during situational or developmental crises experienced by their children. Families may require additional professional assistance at those times to deal with the resultant recurrent grief.

Anticipatory Grief

The extended period of illness experienced by children with NLTIs may provide families with time to prepare for the death of their child. Kubler-Ross (1983) proposed that when families have time to mourn losses as they occur, then the seriousness and frequency of psychological difficulties following the child's death will be minimal. However, as noted by B. Davies and Eng (1993), the value and function of anticipatory grief has not been conclusively demonstrated. One

difficulty with the concept of anticipatory grief is that it leads to the assumption that an expected death is easier to deal with than an unexpected death. Yet, this view may be erroneous, since the expected death of a child appears to have as severe an impact on parents as does a sudden bereavement (Hill, 1994). In addition, many clinicians have observed that, despite education and anticipatory grief, families are rarely prepared for the reality of their child's death. B. Davies (1993) noted that parents and siblings, who have accepted the inevitable outcome of the child's illness, often make comments after the death such as, "But I didn't think he would die until he graduated" or "I thought he would wait until I got home from school"(p.141). It appears that anticipatory grieving may have a role to play in families' grieving. However, that role remains unclear. It cannot be expected, therefore, that anticipatory grieving will reduce the need for assistance among families with a child who has an NETI.

In summary, current theorists are replacing the early conceptions of grief as linear and time-bound with a new conceptualization of grief as an active, dynamic process that may continually evolve yet never reach a conclusion. This newer conceptualization of grief was adopted as a conceptual basis for the current study of families where a child has an NLTI. The concepts of chronic sorrow and anticipatory grief were also included in the conceptualization in anticipation that they may have relevance for working with these families.

Perspective on Children and Life-Threatening Illnesses

Children were central in this study. It is critical, therefore, to consider varying perspectives on children and life-threatening illnesses. Different perspectives will lead to alternative conceptualizations that could, in turn, lead to different research designs and data collection methods. In this study, children were considered active participants in the family. It was assumed that, providing they could communicate, children who were touched by life-threatening illnesses would be capable of thinking about and discussing death and dying.

Children's Knowledge About Death

Early assumptions about how to deal with children who faced life-threatening illness were based on how well children developed their concept of death. Development of this concept was thought to parallel Piaget's sequence of periods of cognitive development (Piaget, 1960). Adults, therefore, thought that death meant separation to infants and toddlers. For pre-schoolers in the stage of preoperational thought, death would then be viewed as both separation and departure. Death would be like sleep, and dead people could feel when people visited their graves. These children would sometimes think that death was reversible. In the period of concrete operational thought, age 6-10 years, death would be seen as real, irreversible, and something that happened to other people. This group would tend to personify death and to think of it in terms of an outside agent. As children got older, and especially with the development of formal operational thought, death would be viewed as the end of life. It would involve irreversible decomposition of the body. In late adolescence, death would also be conceived as the end of personal time, as a cruel personal blow, and as the loss of newly developed intimate relationships (Bartholome, 1995).

Based on the views that were prevalent early in the development of knowledge about children and death, professionals advocated a closed approach to protect children (Stevens, 1993; Waechter, 1971; 1985). Parents were told to maintain a sense of normality, to shield the child from knowing how seriously ill they were, and to remain cheerful and pretend that everything would be alright. However, some clinicians challenged the protective view (Bluebond-Langner, 1978; Spinetta, 1974; Waechter, 1985). They noted that ill children were anxious and knew about their disease. These clinicians observed that children who had the opportunity to discuss their illness, prognosis, and concerns seemed less anxious than children who were deprived of this opportunity. One study of children with life-threatening illness provides support for these observations (Waechter, 1971; 1985). Death anxiety among 64 children between the ages of 6

and 10 years was studied through the use of a projective test and anxiety scale. The children (matched for sex, age, race, social class, and family background) were divided into four groups: children with chronic disease who were expected to die; children with chronic disease, but a good prognosis; children with a brief illness; and non-hospitalized, well children. Only two children with fatal disease had been told their diagnosis and had discussed their concerns about death with their parents. Death imagery was found in 63% of the stories told by all of the children. Most of the children seemed aware of their prognosis despite efforts to protect them from knowledge about their disease. Those children with fatal illness who had more opportunity to discuss their fears and concerns also expressed less specific death anxiety.

Although there are now over 50 studies of children's understanding of death, some inconsistencies exist among studies (Faulkner, 1993). Some of the discrepancies in results are related to methodological variations, such as some studies had small sample sizes or did not include a full age-range of children. In addition, many studies were limited to white, middle-class, suburban children and so, may not be applicable to other children. Difficulties also arise when trying to compare studies using disparate data collection methods including standardized interviews, descriptions of death-related pictures, and spontaneous or directed play. Although standardized interviews were most commonly used, variations in the difficulty and specificity of questions prevent direct comparison of results across studies. Inconsistencies also result from the difficulty in defining the phases that children pass through as their concept of death develops. Although level of cognitive development has been broadly linked to a child's understanding of death in some studies (Koocher, 1974), not all researchers agree (White, Elsom, & Prawat, 1978). Faulkner suggested that part of the difficulty arises because Piaget did not directly examine how children learn about death. Thus, researchers have had to extrapolate which cognitive skills a child needs to understand the components of a mature concept of death.

It appears that information about a particular child's understanding of death can only be obtained by discussing the issue with the individual child (Faulkner, 1993). Evidence suggests that a child's concept of death is related to his or her own experience with illness and its treatment, rather than only to the child's chronological age or stage of cognitive development (Reilly, Hasazi, & Bond, 1983). Children who have experience with death understand death in more depth and at an earlier age than children without this experience (Reilly et al.). In addition, the family's culture and environment, and children's wide variation in individual rates of development (physically, emotionally, cognitively, and intellectually) are also said to affect the child's developing concept (DeSpelder & Strickland, 1996).

Communicating With Children About Death

Professionals need to understand how a child's approach to death is related to developmental capacities, as well as to innate differences and past experiences. Even children as young as four years of age may be aware of the seriousness of their illness. In recent times, families tend to be more open in their communication, although actually discussing death may still be avoided. Goldman and Christie (1993) studied children over three years of age and their families (N=31) to determine whether they discussed the child's impending death together. Twenty six of the children had been told that their disease had recurred. In 28 families with siblings over three years of age, 75% had told the siblings about the impending death. Goldman and Christie also interviewed staff members (N=22) in the oncology unit about their attitude to an open approach in talking about death and dying and how often it occurred. Factors that influenced communication included the length of time the child had been ill and the family's communication style. Children who had been ill the longest were more aware of the severity of their illness. Each family with an open communication style acknowledged the impending death. All staff agreed or strongly agreed that open communication is the preferable approach. Some qualified it, however, with the proviso that parents should also be happy with this approach. Staff

thought that 10-80% (median of 45%) of children talked about their own death with their families. A lack of communication skills on the part of both staff and families may make it difficult to discuss death with the ill child. Additionally, maintaining openness may become increasingly difficult with relapse and impending death.

Despite what many people think, children are not miniature adults. A child's reality is different from that of an adult. Children do not understand the world from an adult's perspective (Bartholome, 1995; Faulkner, 1993). They do not have a well-developed sense of self. Moreover, they live in a world where fantasy and reality are not well divided. Children cannot understand the concepts of chance and probability, so everything has an immediate and proximate cause. They are also egocentric. They relate everything that happens around them to their own feelings, ideas, and actions. Children may use concretistic thinking to sort out their world. They have their own sense of time. They have no sense of physical or personal time. Time is not a linear progression, but is collapsed into the present (Bartholome). Children also listen more to what adults do than to what they say, so non-verbal language is important. In turn, professionals and families need to look for cues to what the child wants to talk about. Moreover, they must be prepared to accept fears and anxieties related to loneliness, mutilation, and death that seem irrational from an adult perspective, because the fears will be quite real to the child (Faulkner).

Based on clinical experience and available research, Stevens (1998) provided guidelines for caregivers of seriously ill and dying children. According to Stevens, these children know a lot about their situation, so it is ineffective and damaging to conceal information from them. Questions should be answered truthfully, especially since the child often knows the answer and is just seeking confirmation. Children will often accept information about their prognosis in a matter of fact way. Those who do not ask questions may be trying to protect their parents (Bartholome, 1995; Stevens). Infants who are too young to have a concept of death still need physical relief and comfort. Young children may have difficulty in expressing their fears of

separation. They will respond to the anxiety around them. Discussing fears and promoting family togetherness is believed to be important for children of 2-7 years. Expressive play with art and music may be helpful in encouraging children to share their fears and feelings (Gray, 1989). Children of 7-12 years of age may fear abandonment, destruction, and body mutilation. It is suggested that they need people to be truthful with them and to foster their sense of control over their deteriorating body. Friends are also important and should be encouraged to visit. One of the tasks of adolescence includes separation from parental control. Adolescents often struggle with the paradox of wanting support and yet wanting to meet challenges by themselves. They may prefer to confide in a peer group, especially with others in a similar situation. As with younger children, adolescents may be more concerned about their family and friends than about themselves. They tend not to be afraid of death so much as afraid of dying. Professionals are encouraged to listen carefully to understand the adolescents' perceptions of their own illness and prognosis. Privacy and a sense of independence is important to an adolescent, as is peer contact and support. Offering choices may give adolescents a sense of being in control, and may reduce anger, frustration, depression, and anxiety. Writing poems, letters, or a journal or drawing can release pent-up emotions. Many adolescents want to know that they will be remembered. Creating a permanent record to leave behind, such as video, tape, or photograph, may be valuable (Stevens). While these guidelines are helpful, further research is needed to evaluate the efficacy of these approaches.

Siblings.

Surviving siblings are particularly vulnerable. They should not be forgotten (Gibbons, 1992). They are also children and so their reality is that of a child. Consequently, siblings may have their own version of what caused the child's illness. They may have misconceptions about the nature of the illness or about the hospital clinic and treatment program (Sourkes, 1980; Stevens, 1998). Sourkes also found in her study that siblings may have a fear of developing the

same illness. Yet at the same time, they may feel guilty and ashamed because of their relief that they do not have the illness. In addition, siblings may have ambivalent feelings about the ill child, such as envy or shame. They may be so stressed by the illness that their schoolwork and relationships suffer (Sourkes; Stevens). B. Davies and Martinson (1989) reported from their research with siblings of children who had died of cancer that emotional realignment within the family, family separation, and the ill child's therapeutic regimen can be very stressful for siblings. These stressors may result in negative consequences such as feelings of loneliness, resentment, rejection or neglect, frustration, fear, and sadness. Some clinical observations suggest that parents may be unable to respond to siblings for a variety of reasons (Gibbons). Parents may be unavailable or detached, may cease family celebrations, may feel inadequately prepared to discuss death with siblings, may think siblings are too young to understand, or may want to protect siblings. Moreover, siblings may interpret a lack of communication as an accusation that somehow they are at fault. The lack of communication may isolate siblings even more. Some parents may become overprotective of siblings. Or, they may withdraw from parental involvement because they are afraid to lose this child too (Gibbons).

B. Davies (1988) reported that the family is critical in creating the environment in which siblings respond to the death of a child since children are dependent on their parents (and adults) for information and support. They often do not have any other source of help. In her study of 34 families 2-36 months following the death of a child, B. Davies used semi-structured interviews and standardized instruments to examine family environments. She discovered that siblings in the more cohesive, active, and religious families had fewer behavioural problems. B. Davies suggested that families who participated more in social activities were promoting adaptation in siblings. Social support was also noted as important for the siblings. B. Davies suggested that professionals could contribute to sibling adaptation by encouraging and facilitating family cohesion, having information sessions with the family and not just with the parents, asking

family members to assist in the care of the dying child, and treating the patient and the family as one unit for care.

In conclusion, professional understanding of how children develop their concept of death has changed over the years. It is now recognized that a child's perception of reality and concept of death is not the same as an adult's. Some theorists and clinicians recognize the importance of open communication instead of refusing to discuss death. Siblings are also children. They are vulnerable when a child is dying and should not be forgotten. Clinical observations and available research have provided an important foundation for the development of guidelines to assist professionals in supporting ill children and siblings (Stevens, 1998). Few of these recommendations, however, have been adequately evaluated.

Perspective on Families

Family means different things to different people. There are numerous definitions of family, such as those that emphasize relationships through blood ties, adoption, guardianship, or marriage. Other definitions focus on social or emotional relationships between at least two people in a single household. Earlier definitions of family reflected the traditional nuclear and extended family. Definitions have become broader in the past 10-15 years, and now include a variety of possibilities, such as single-parent families, blended families, and same-sex families (Friedman, 1998; Hanson & Boyd, 1996). In this study, the family was viewed as a complex unit that is composed of individual members, but that has distinct attributes of its own. Ideally, the family would be self-defined and include whoever the family says it includes. The family unit in this study, however, was limited by the researcher to those members with whom health care professionals most often interact and who usually play a key role in pediatric palliative care - the ill child, parents, and siblings.

There are also several different approaches to family nursing. These perspectives include the family as the context for individual development; the family as the unit of care; the family as

a system; and the family as a component of society (Friedman, 1998; Hanson & Boyd, 1996). In this study, the family was viewed as an interactional system in which the whole is more than the sum of the parts. It was assumed that systems data are needed in order to provide care to all family members, as well as to the family as a unit (Feldstein & Rait, 1992; Friedman). Family systems theory provided a useful guide for examining families' interactional relationships (Friedman; Jassak, 1992), and for directing the process of data collection. The following overview of systems theory further explicates the perspective on families that formed part of the context for this study.

Systems Theory

Systems theory explains the behaviour of complex, organized systems. It is a way of looking at the world in which objects are interrelated with one another. von Bertalanffy (1968; 1975) recognized living systems as a complex organization of many parts. He sought to account for the seemingly animate behaviour of non-living systems in terms of complex inter-relationships and transactions among components. Holism is a key feature in systems theory. A system must be understood as a whole. It cannot be comprehended by examining its individual parts in isolation from each other. There are properties of the system that do not derive from the components and only emerge at the systemic level. Human systems are self-reflexive. Thus, they can examine their own system and set goals for themselves. Communication is extremely important and humans behave according to the meaning a thing has for them (von Bertalanffy).

Transactional patterns, rather than individual responses, are of interest in families. Family processes can be understood as a product of the entire system, instead of any one person. These overall patterns of interaction within families, rather than each separate member's behaviour, illuminate family processes and reflect the family system (Forchuk & Dorsay, 1995). When one examines family systems within a hierarchy of systems (subsystems are smaller than the family unit and suprasystems are larger than the family), then one can explore questions in more depth

and can explore how family systems change. Morphogenesis is the process of family change by the whole family system, rather than by just a single family member. First-order change is when individual behaviour changes without the completion of a positive feedback loop. Whereas second-order change is a higher more major level of change that can alter the system itself.

A system is a set[s] of elements standing in interrelation among themselves and with the environment (von Bertalanffy, 1968; 1975). A system, therefore, is either a set of interrelationships or a set of components, depending on the level of analysis. In family research, individual family members are the components. They are interdependent, and behaviours of the components exhibit mutual influence. That is, each member's behaviour affects every other member of the family. Hierarchies and the issue of power are important in families; for example, a parental subsystem may exert more power than an offspring subsystem.

Systems require inputs, throughputs, and outputs that are interrelated by rules of transformation that take place in systems. Families may become immobilized because they do not have a family rule to use in dealing with a particular situation, such as when a child is dying. Equifinality is the ability of a system to achieve the same goals, but through different routes. Goal-oriented behaviour results from communication of information among components and from how this communication is organized, especially through feedback loops (Friedman, 1998). Feedback loops are closed circuits or paths in which information goes from one part, through others, and back to the origin. A negative loop operates to restore or maintain equilibrium (homeostasis), while a positive loop increases deviation from the status quo. If the structure of the system stays the same, then it is *morphostatic*; if the structure changes, then it is *morphogenetic* (von Bertalanffy, 1968; 1975). Although overall control of the behaviour of the system as a whole does not reside in one component, a single component can become increasingly important in determining the behaviour of the whole (*progressive centralization*). Thus, a dying child may exert unusual influence on the family.

Boundaries.

Boundaries are an essential concept in systems theory. If one can identify components of a system, then one is really drawing boundaries around them. A boundary defines the system and marks the interface between the system and its environment. Human systems are embedded in the environment, so one can only gain understanding of families if one takes into account the sociocultural, historical, political, and economic contexts (Friedman, 1998). Boundaries can be determined by persons inside or outside the system. However, there may not be consensus about the boundaries because determining a boundary requires self-reflexivity and some degree of judgement. Systems are said to be open or closed depending on their boundaries. Boundaries are impermeable (closed to interchange from outside the system), semi-permeable (allowing some interchange), or amorphous or transparent (offering no impediments to interchange). All families are open systems, although in varying degrees. One needs to determine what or who permeates the boundaries. In many family assessments, boundaries are operationalized as: (1) degree of permeability or, from another perspective, of cohesiveness; and (2) emotional connectedness among family members (Klein & White, 1996). The rigidity of the family structure affects how smoothly families can adjust to entrances and exits across the family boundaries. When there is boundary ambiguity, such as might occur as a child is dying, then uncertainty about who is in or out of the family system may be immobilizing. Whether one believes that it is the person per se or their roles in the system that are important to the family system makes a difference to the meaning of a family member's death. If one believes in the importance of roles, then someone else will be able to fill the empty role. However, if the individual person is more important, then no one can fill the empty place.

Family Research

In pediatric palliative care, the unit of care is the family and not just the individual child. Clinicians and researchers recognize that patients exist within a family system and also within a

myriad of social systems (Clarke, 1995; Friedman, 1998; Gulla, 1992). Until recently however, families have been neglected in health care research. There is little research and guidance available to assist health professionals in providing family-level care (B. Davies, Reimer, & Martens, 1994; Friedman). While family-focused care often implies that health professionals must view the family solely as a unit, it is important to note that one cannot view the unit as a whole without giving consideration to the individuals who make up the unit (B. Davies, Reimer, Brown, & Martens, 1995; Friedman).

The definition of family must be explicit in family research. Further, articulation of a conceptual framework guides research design and methodology decisions (Boyd, 1996). Data from families should be collected and analyzed in a manner that is consistent with the researcher's conceptualization of the family (Copeland & White, 1991; Gilliss & Davis, 1992). In this study, family was defined as a complex unit composed of individual members, but with distinct attributes of its own. Accordingly, data needed to be collected from individual members, as well as from the family as a unit. In addition, analysis had to take both the individual and the family unit perspectives into account.

Family researchers have primarily focused on individuals, rather than the entire family unit (Broome, 1998; Leahey & Wright, 1987). The underlying assumption has been that if one knows an individual's role, such as mother or father, then one can determine the behaviours of the unit (Gilliss & Davis, 1992). Researchers have tended to make inferences about the family based on only the mother's experiences (Birenbaum, 1995; Kristjanson, 1992). While Birenbaum suggested that, at times, one member of the family may be the best informant about a family phenomenon, relying on reports of a single individual or subset has the potential to present a biased view of the family when the family unit is the focus of study (Lynn, 1995). In this research study, the views of the individual members and the family as a whole were all considered significant. Therefore, all members of the family were encouraged to participate to

the maximum extent possible, recognizing that participation was restricted by age, developmental stage, and cognitive ability.

Obtaining accurate information from the family about the family can pose a problem for researchers. The information obtained from each source of data may be different and there may be a lack of agreement among the family members. One may find a lack of correspondence between individual reports and those of the family as a whole. However, although one may get different views, they may be complementary to one another. Uphold and Strickland (1989) suggested that obtaining information from multiple members of a family is beneficial and performs a kind of triangulation that gives a better picture of the whole. Triangulation, either using multiple data sources or multiple methods to obtain data, may improve the researcher's ability to find out about the families' experiences (Woods & Lewis, 1992). Both participant observation and interview methods of data collection were used in this current study. Sources of data also included individual family members as well as the family as a group.

A criticism of this approach centres around the meaning of triangulation and the implicit expected outcomes. Triangulation implies that one is focusing on a central point, in this case "the family experience," in an attempt to discover "the truth." However, there may be multiple "truths" within any family. The experiences of less powerful members of the family, such as children, have tended to be ignored in the past. Although involving children may require creative methods of data collection such as the use of drawings or poems, they are able to and should be encouraged to contribute. Family research needs to value the contribution of all members of the group, and to view their perceptions of the situation as being equally relevant (B. Davies et al., 1994). If different members report different "facts," the researcher should not then decide which person carries more weight and is more "truthful" in representing the family experience. Instead, as in this study, specific "facts" should be viewed as part of the data.

Research examining the experience of the family as a whole must reflect the dynamism in family systems, and must use measures that portray families as entire units (Broome, 1998; Ferketich & Mercer, 1992; Woods & Lewis, 1992). However, when one's level of inquiry involves the family as more than the sum of its parts, no adequate tool exists to measure the whole. It has been noted that though family theorists and therapists are concerned with family systems and family psychopathology, no widely accepted concepts of relational and systemic pathology have yet been developed (Cowan, Cowan, & Schulz, 1996). Complex systemic models that combine conceptual, measurement, and statistical techniques require further investigation (Cowan et al.). In quantitative research particularly, questions are raised about how to combine individual family member data into scores that reflect the family as a unit (Clarke, 1995; Ferketich & Mercer; Uphold & Strickland, 1989). For example, is a summative score derived from individual scores an accurate representation of a family's score? Can one obtain a score about a family simply by aggregating the individual family member's score on a given instrument? Is the maximised family score a good indicator of impacts on the family as a whole? What value is there in highlighting differences in scores between family members as the central concern of a study? These and similar questions raise issues that continue to be debated. There are unique weaknesses and strengths in each approach (Uphold & Strickland). Currently, no adequate answers exist to these questions. Again, one must base the choice of sources of data on the purpose of the study, the theoretical basis for the study, and the population of interest.

Since the definition of family and its boundaries is often artificially and externally imposed and may not reflect the boundaries that would be meaningful to the family, Kristjanson (1992) suggested that research based on families' own definitions of their membership may be useful for some research questions. Moreover, family research is transactional, thus one should examine patterns of family behaviours (Kristjanson). However, the context of the data collection must also be taken into account because individuals may respond to the same question in

different ways if asked individually or in a family group (Astedt-Kurki & Hopia, 1996; Kristjanson). Birenbaum (1995) suggested that grounded theory methodology, involving data collection from both the individual family members and from the group as a whole, is an efficient way of collecting and analyzing family-level concepts. Grounded theory, involving data collection from both individual family members and from the family as a whole, was the method chosen for this study.

Families differentiate and carry out their functions through subsystems within the family, such as spousal, parental, and sibling subsystems. Loss, actual and potential, is a major transition that disrupts patterns of interaction and requires family reorganization (Martinson, McClowry, B. Davies, & Kuhlenskamp, 1994; Walsh & McGoldrick, 1988). "Off-time" losses, such as the death of a child, are often viewed as the most painful ones. However, although members of a family may be affected by the terminal illness, one cannot assume that all members are adversely affected (Birenbaum, 1995). Members of the family are often not affected in the same way, nor do they respond in a similar fashion. Birenbaum called for research to identify who is affected and why, because some individuals seem capable of buffering themselves against an event. Within a family, the unique constellation of relationships probably affects the impact of the death on the family members and on the family as a whole. Therefore, one must examine the relationships pertaining to each family member, as well as examining the family group as a whole and distinct entity.

Families do not exist in isolation from the larger community. Particularly as a child's illness progresses, families may require increasing external help and support (Jefidoff & Gasner, 1993). Families must then seek treatment, information, and services for the child and themselves outside of the family, while concurrently managing these systems. For example, families in Jefidoff and Gasner's study needed to discuss their fears about the child's death. Yet, they also needed to prepare other family members and friends for the imminent death. Clinicians have

observed that the interaction patterns of family members, their interactions with others outside the family system, such as friends and neighbours, and the degree of social support may influence a family's ability to cope with the experience of having a terminally ill member (Ross-Alaolmolki, 1985). The interaction between the family system and the health care delivery system may also be important. It may have a direct bearing on how patients and families come to understand their roles during illness (Speedling, 1983). Hospital norms and regulations may implicitly and explicitly affect the family's access to information. The attitudes of personnel may influence the family's ability to develop competence in performing the required roles. Larger systems, such as hospital wards and physician-family relationships, may foster dependency on the medical system and preoccupation with caregiving. Thus, families may be prevented from developing new and flexible roles (Rosman, 1988). Speedling suggested that, "Observing families as they live through the process [of illness] can provide vital information about what they need at different stages of the process and how the current system for delivering care affects the quality of coping" (p. 75). The current study used this type of observation to uncover such information.

To summarize, little research has been from the perspective of the whole family. Most research has been retrospective and has involved only the parents, and sometimes only the mother, to provide a family perspective. Yet, children perceive the world differently from adults. Thus, the children's perspectives must also be sought. Moreover, the literature is unclear in offering guidance to assist health professionals in providing family care and conducting family research. A central issue in family research concerns how individual reports on experiences can be used to provide insights about the family unit. Recently, researchers have suggested that the views of all family members should be treated as equally important, and differences between accounts should be viewed as further data for a higher level of analysis.

Perspective on Coping

The individual coping literature is well established, but family coping remains a developing concept (Birenbaum, 1991; Cowan et al., 1996). Family coping is complex and difficult to measure. For example, the members of a family who participate in the family's coping process are at various developmental levels. How, then, can one use similar data collection methods when family members may range in age from infants to the elderly? Similar issues have been raised about developing instruments to assess family coping. For example, can one describe family coping simply by aggregating the individual family member's styles of coping scores? Can issues of power within families be accounted for so that children have a voice? Or should responses be weighted to reflect the status of individuals within the family?

Despite the emphasis on family as the unit of analysis, the perceptions of individuals continue to be the focus of family coping measurement (Birenbaum, 1991; Broome, 1998; Cowan et al., 1996). Most are designed as individual, self-report instruments to assess perceptions about the family as a unit (Kristjanson, 1992). Thus, they provide relational-level data that are not necessarily meaningful when exploring interactional relationships (Sullivan & Fawcett, 1991). In addition, family coping instruments are often just administered to parents (Birenbaum). The role of children in family coping, therefore, is often ignored. Because of the continuing use of individuals to represent the family, it is important to identify what is known about individual coping. This section will begin with examination of one model of individual coping that dominates current theorizing in the field of stress and coping. Limitations of this model for the current study will be identified prior to examining a family model that is prominent in the existing family coping literature.

Individual Coping

The transactional model of coping proposed by Lazarus and Folkman (1984) is a model of individual coping that is well known in health care and in the stress field. In this model, two

processes, cognitive appraisal and coping, mediate the person-environment relationship. These processes then determine adaptational outcomes. Cognitive appraisal determines whether and to what extent a particular transaction or series of transactions between the person and the environment is stressful. There are three types of cognitive appraisal: primary appraisal; secondary appraisal; and reappraisal. Primary appraisal allows the person to classify the transaction as irrelevant, benign-positive, or stressful. Secondary appraisal involves the evaluation of what might and what can be done. Secondary appraisal takes into account which coping options are available, the outcome expectancy, the efficacy expectancy, and the consequences of using a particular strategy. Reappraisal is "...a changed appraisal on the basis of new information from the environment, which may resist or nourish pressures on the person, and/or information from the person's own reactions" (Lazarus & Folkman, p.38). Reappraisal indicates that appraisal of the situation may change over time.

Many factors affect cognitive appraisal. Person and situational variables are interdependent. Among the most important person factors are commitments and beliefs. They help to determine the saliency of an encounter in relation to well-being. They also shape the person's understanding of an event and, therefore, his or her coping efforts. Commitments also influence appraisal through their impact on vulnerability. Lazarus and Folkman (1984) state that, "The greater the strength of a commitment, the more vulnerable the person is to psychological stress in the area of that commitment" (p.58). Beliefs about personal control and existential beliefs are particularly relevant to appraisal. Situational variables are interdependent. Together with personal variables, they contribute to an appraisal of harm/loss, threat or challenge. Situation variables include novelty, predictability, event uncertainty, temporal factors, ambiguity, and timing of the stressful events in relation to the life cycle (Lazarus & Folkman).

Coping is affected by cognitive appraisals. Lazarus and Folkman (1984) define coping as "...constantly changing cognitive and behavioural efforts to manage specific external and/or

internal demands that are appraised as taxing or exceeding the resources of the person" (p.141). Coping functions are emotion-focused or problem-focused. These functions may occur simultaneously, and may either facilitate or impede the coping outcomes. Coping is a process that implies effort. No strategy is better than another one. When judging effectiveness, consideration must be given to the context and the frame of reference. Effective coping must be judged from the client's perspective, rather than only from the researcher's opinions or values about how the person should be coping. Effective coping is not equated with mastery over the environment. The dynamics and the change that characterize coping as a process are the function of the continual appraisals and reappraisals of the changing person-environment relationships.

The major adaptational outcomes are social functioning, morale, and somatic illness. Each outcome can be conceptualized as having both a short-term and a long-term component, with short-term outcomes reflecting coping with a specific encounter. Relationships between the short- and long-term outcomes are complex in that the nature of the short-term outcomes are not necessarily associated with similar long-term outcomes. The relationships between the three major adaptational outcomes are also complex. Good functioning in one area may be at the expense of functioning in another area (Lazarus & Folkman, 1984).

The basic assumptions of this transactional model of coping (Lazarus & Folkman, 1984) maintain that coping is effortful on the part of an individual; is situationally focused; and is accessible to the consciousness, so can be reported. This model, therefore, can be used to describe an individual's response to an acute episode or event. Families who are living with a child with an NLTII may face acute episodes, but they also face persistent demands that are part of their daily lives. How, then, can a set of behaviours and cognitions be described as coping, as opposed to ordinary living? Moreover, Gottlieb (1997) suggested that coping associated with recurrent or persistent life difficulties may be more or less effortful or automatic depending on when questions are asked. Coping efforts eventually become incorporated into one's daily life.

They are no longer associated with a specific stressor. Individuals, therefore, may not be aware that they continue to use a particular way of coping. Consequently, the Lazarus and Folkman model has limited applicability in research with families living with a child who has an NLTI. First, reliance on the individual's perspective fails to explore the family's perspective. Second, coping is only part of a family's life. Using this framework would restrict the scope of the study. Thus, important aspects of families' lives may be overlooked. Finally, the possibility that individuals routinize some coping efforts, and so are unable to consciously identify them, suggests that the researcher may disregard or fail to uncover certain coping efforts employed by families if the focus is on conscious coping efforts.

Given that individual coping has not been shown to reflect a family process, individual coping frameworks were not viewed as appropriate for this current study. In keeping with a family systems view of the family, theoretical frameworks about family coping that are based on systems theory were reviewed. The tenets of one major family coping framework are discussed in this section to highlight its strengths and limitations for this study.

Family Coping

The Resiliency Model of Family Stress, Adjustment, and Adaptation is a family coping framework that is widely used in health care (McCubbin & McCubbin, 1993). This model builds on the Double ABCX model of family stress theory developed by McCubbin and Patterson (1982), which in turn was expanded from the original family stress theory proposed by Hill (1949). In earlier renditions of McCubbin and Patterson's model (1982), "A" denoted a stressful event with associated hardships; "B" represented the family's physical, psychological, material, social, spiritual, and informational resources; "C" demonstrated the family's subjective definition of the stressful event; and "X" denoted a crisis, and the amount of disruptiveness or incapacitation within the family due to the stressful event. The models are based on a number of assumptions: (1) the family is a system; (2) unexpected or unplanned events are perceived as

more stressful than other events; (3) stressful events within the family are more disruptive than those outside of the family; (4) lack of previous experience with a stressor leads to greater perceived stressfulness; and (5) ambiguous stressor events are more stressful than nonambiguous ones.

In the Double ABCX Model, the "C" factor was expanded to "CC" and was defined as the family's perception of the original stressor and of the pile-up of other stressors ("AA"), plus the family's perceptions of its resources ("BB"). The concept of coping was incorporated into the Double ABCX Model and was perceived as an adaptive process in families wherein resources, perceptions, and behaviour interacted (McCubbin & Patterson, 1982). Coping included both cognitive and behavioural strategies. Altering the meaning of the situation was viewed as one way in which families coped with situations where demands exceeded capabilities. In addition, a more generalized meaning construct was introduced to the Double ABCX Model. This construct was a sense of coherence (SOC) which was based on the salutogenic model of health proposed by Antonovsky (1979) in answer to the prevailing pathogenic model. Antonovsky suggested that it is important to examine how people successfully cope with stressors and what influence that successful coping has on health. He developed the SOC and defined it as a global orientation which sees the world as more or less comprehensible, manageable, and meaningful. According to Antonovsky, the SOC is a determinant variable that affects health consequences and other aspects of well-being.

The Double ABCX Model was eventually renamed the Resiliency Model of Family Stress, Adjustment, and Adaptation to emphasize adaptation as the central outcome of the stress process (McCubbin & McCubbin, 1993). This emphasis on potential positive outcomes was consistent with the influence of Antonovsky's work (1979; 1987) on individual and family resilience, and the results of other researchers' studies of resilience. Two levels of meaning were differentiated. Situational meanings were defined as the individual's and family's subjective

definitions of demands and capabilities, as well as the relationship between them. Global meanings were viewed as transcending any given situation and reflected a more stable set of cognitive beliefs about the family's relationship to society and the relationship among family members. The goals of the family unit's behavioural responses were to eliminate stressors, manage hardships, resolve intrafamilial conflict, and develop resources to facilitate family adaptation over time (McCubbin & McCubbin).

There are two phases in the Resiliency Model (McCubbin & McCubbin, 1993). The Adjustment Phase starts with an illness stressor that interacts with a family's vulnerability. This family vulnerability to life changes and pile-up of stressors interacts with the family's type and established patterns of functioning. These components then interact with the family resistance resources, such as the quality of communication between husband and wife. This, in turn, interacts with the family's appraisal of the illness stressor and its severity. Finally, interaction continues with the family's problem solving and coping strategies. The result of these interactions is either good adjustment with positive outcomes, or a maladjusted crisis situation. Family crisis may be viewed as an expected outcome of the numerous stressors that often accompany chronic illness. It requires a change in patterns of family functioning that denotes the beginning of the Adaptation Phase of the Resiliency Model (McCubbin & McCubbin).

The adaptation phase is also determined by a number of interacting components. The pile-up of stressors ("AA") interacts with the family's resiliency that is partly determined by the family's type and newly instituted patterns of functioning. These components interact with the family's resources and capabilities ("BB") which are supported by family and friends ("BBB") and the family's appraisals. The family's situational appraisal ("CC") then interacts with the family's schema appraisal ("CCC") and creates meaning. Finally, the resource and appraisal components interact with the family's problem-solving and coping strategies to facilitate adaptation. Family adaptation is the outcome of efforts to achieve coherence and functioning in a

family crisis situation. These efforts are directed at achieving balance and fit at both the individual-family level and the family-community level. Some families will become well-adjusted over time, while others will remain in a maladjusted crisis situation ("XX").

According to McCubbin and McCubbin (1993), families are more likely to adapt successfully to an illness stressor if they are less vulnerable because fewer other stressors or changes are occurring concurrently; if they have more adaptive patterns of functioning, such as more emotional closeness and more flexibility in roles, boundaries, and rules; if they define the situation positively and feel they can master or control it; and if they have good coping and communication skills. Further, a family's SOC is a mediating coping factor between family stressors and adaptation. The higher a family's sense of coherence, the more likelihood of successful adaptation (McCubbin & McCubbin).

The Resiliency Model of Family Stress, Adjustment, and Adaptation (McCubbin & McCubbin, 1993) is a model that is undergoing continual revision in response to research evidence. It provides a somewhat useful framework in which to examine family experiences of a child's progressive, life-threatening illness. However, the framework has some significant limitations for this population that must be addressed. First, most of the research on the model has been done with healthy families (Danielson, Hamel-Bissell, & Winstead-Fry, 1993). Recent research has begun to examine families where a member has a disability or chronic illness (Patterson & Leonard, 1994; Patterson, Leonard, & Titus, 1992), but there is no research on progressive, life-threatening illnesses. In addition, since there is very little research at all in pediatric NLTIs, one must be open to discovering the potential scope of families' experiences. The use of a theoretical framework may bias researchers and may prevent or restrict them from seeing the richness and the full extent of families' experiences when a child has an NLTi. Second, the model is based on enhancing understanding of family adjustment and adaptation responses to stressful situations. While this goal is laudable, the emphasis on responses to

stressors and coping strategies has limited meaning when one is trying to understand a family's experiences of living with a child who is dying. Understanding experiences encompasses coping, but also includes the fuller and richer description and theoretical formulation of family life. Finally, one of the major concepts in the model, the resiliency concept of SOC, is based on research with individuals. Family global meanings have not yet been operationalized and tested. It was, therefore, premature and constraining to use the Resiliency Model as a framework in this current study which examined the experiences of families as a whole, rather than the perception of only one family member.

Summary

The conceptual framework for this study was based on particular perspectives on children and life-threatening illnesses, families, and coping. The focus of this study was where these three perspectives intersected within the broader context of grief. Grief was viewed as an active, dynamic process that may continually evolve, yet never reach a conclusion. This more recent understanding of grief challenges professionals to reconsider the needs of families when a child has an NLTI. In addition, the kind of interventions that will be most helpful should also be reconsidered. The concepts of chronic sorrow and anticipatory grief were also considered potentially relevant to families where a child has an NLTI.

Children, both ill children and siblings, were considered active participants in the family. It was assumed that, providing they could communicate, children who were touched by life-threatening illnesses would be capable of discussing issues related to death and dying. Open communication, rather than hiding death, was considered important. Professional understanding of how children develop their concept of death has changed over the years. It was recognized that a child's perception of reality and concept of death is not the same as an adult's. The vulnerability of siblings when a child is dying was acknowledged.

In this study, systems thinking informed the perspective on families. The family was viewed as a complex unit that is composed of individual members, but that has distinct attributes of its own. The unit of care was seen as the family and not just the individual child. The unit as a whole could not be viewed without considering the individuals who made up the unit. The contribution of all members of the group was valued. Their perceptions of the situation were viewed as being equally relevant. This perspective on family guided research design and methodological decisions. Little research has been conducted from the perspective of the whole family. Most family research has been retrospective and has involved only the parents, and sometimes only the mother. Currently, there is little research and guidance available to assist health professionals in providing family-level care.

The final concept underlying this study was coping. The individual coping literature is well established, but family coping remains a developing concept. Family coping is complex and difficult to measure. Despite the emphasis on family as the unit of analysis, the perceptions of individuals continue to be the focus of family coping measurement. Neither individual nor family coping frameworks were perceived as appropriate for this current study. The emphasis on responses to stressors and coping strategies had limited meaning because it was considered that understanding family experiences encompasses coping, but also includes a fuller and richer description and theoretical formulation of family life.

Chapter III: Literature Review

Some researchers (Davies, 1996; Martinson, 1993; Parker, 1996; Stein, Forrest, Woolley, & Baum, 1989; Stein & Woolley, 1990) have begun to investigate the impact on families of caring for a child with a progressive, life-threatening illness (PLTI) at home. However, existing research in pediatric palliative care is minimal. There is little known about families' experiences when a child has a PLTI. Even less has been written about experiences with a neurodegenerative, life-threatening illness (NLTI). Only two studies were found that focused specifically on families of children with neurodegenerative disorders (Davies; Parker). Given the paucity of specific research, the literature review was expanded to include research in the related area of family experiences when a child has a chronic illness or disability. It was assumed that there are some similarities between these families and families where a child has a PLTI. For example, families of chronically ill children also provide most of their child's care at home. An important difference, however, is that families of children with PLTIs must prepare themselves for their child's death. Moreover, the ill child and siblings must also deal with this actuality. Only limited direction, therefore, can be taken from the extensive literature on chronic illness and disability. The available research does suggest that a child's illness has a profound impact on every dimension of family life. Families may be affected emotionally, physically, financially, and spiritually. Moreover, their structures and ways of organization may be permanently altered.

The numbers of children with a prolonged terminal illness are low when compared with those in the adult population. There were, for example, 346 children with terminal illnesses who died in British Columbia over the three year period 1988-1990. At any one time, there are over 200 children in the province who are living with progressive, life-threatening illnesses (B. Davies, 1992). In Ontario, a home-based, pediatric palliative care program established in a major teaching hospital only admitted an average of 16 patients a year over a period of eight years (Kopecky, Jacobson, Joshi, Martin, & Koren, 1997). Yet, one estimate from the United Kingdom

indicates that 1:1000 children are affected by life-threatening illnesses (Goldman, 1996). Approximately 20% of these children have cancer, but many have progressive neuromuscular or neurodegenerative conditions that will eventually cause their death, though not necessarily within a few months or even a few years (Ashby et al., 1991; B. Davies & Howell, 1998; Goldman). Families, therefore, provide care for extensive periods of time. Although the current numbers are relatively small, these children often pose substantial management challenges (Chambers, Oakhill, Cornish, & Curnick, 1989; Hain, Patel, Crabtree, & Pinkerton, 1995; Hunt & Burne, 1995; Hunt, 1990; Kopecky et al.), and may use a disproportionate amount of in-patient health resources (Caring Institute of the Foundation for Hospice and Home Care, 1987). Additionally, the numbers of children with a PLTI are projected to increase as the incidence of life-threatening diseases rises and as advances in technology and medicine reduce mortality rates for conditions from which children previously died (Broome, 1998; B. Davies & Howell).

In this chapter, the literature is reviewed to provide an overview of what is known about families' experiences when a child has a PLTI. First, the potential consequences of living with a child who has a PLTI are detailed to illustrate the possible impacts of an NLTI on families' experiences. Second, factors that may influence the experience are examined to provide a context for understanding families' experiences. Research from the extensive chronic illness and disability literature is used to supplement findings from pediatric palliative care studies. Gaps in knowledge are identified, especially in relation to neurodegenerative illnesses. Particular attention is paid to the experiences of families at home because most of the care for these children occurs at home.

Impact of Families' Experiences

There is limited knowledge available about the potential consequences of living with a child who has a PLTI. The impact of families' experiences has not been well documented. Indeed, the majority of available research only appeared in the literature within the past few

years. This beginning research tentatively suggests, however, that families' experiences have five major impacts. Living with a child who has a PLTI may affect a family's structure and patterns of interaction and may cause emotional, physical, financial, and spiritual disruptions. Research in each of these areas is presented to provide an understanding of the potential impacts of families' experiences when a child has an NLTI.

Impact on Family Structure and Patterns of Interaction

A child's life-threatening illness may disrupt family patterns of interaction, require family reorganization, and pose shared adaptational changes. Some researchers have found that changes in family roles become necessary in order to manage the child's illness (Clarke-Steffen, 1997; Gravelle, 1997; Stein et al., 1989; Stein & Woolley, 1990). Clarke-Steffen interviewed mothers, fathers, ill children, and siblings in a longitudinal, grounded theory study of seven families in which a child was recently diagnosed with cancer. Two semi-structured interviews were completed with each individual, then the family was interviewed as a group in the final interview. She reported that family roles were drastically altered in some families. Some role changes were planned, but many others occurred spontaneously. Typical role changes included mothers changing jobs or stopping work to stay at home with the ill child. Fathers were more likely to rearrange their schedules or take time off work when needed. The bulk of the ill child's care fell to mothers, while fathers often took responsibility for the siblings and increased their household chores. Siblings were sometimes asked to spend more time on the ill child's care and to take on more responsibilities in the household. Similar findings were reported by Gravelle who used a phenomenological approach in her study with eight families of children with a range of PLTIs. Eight mothers and three fathers participated in an unstructured interview, and five of them completed second interviews. Mothers gave up work, sometimes unwillingly, to care for the child. Some felt that they had no other choice. Mothers who assumed their new role unwillingly were often resentful and angry. Stein and Woolley also reported that mothers were

usually the primary caregivers. Although fathers generally continued working, they often used vacation time or took time off work to attend to the child's needs. Some mothers felt particularly housebound because of exhaustion, concern about leaving the child safely in someone else's care, and worry about finding suitable child care. Moreover, these parents also feared leaving their children in case they died unexpectedly. Siblings were sometimes expected to assume extra responsibilities. Consequently, siblings had less free time and fewer friends than their peers (Stein & Woolley).

In many families, the daily routines of family life may be disrupted because families need to follow routines imposed by the child's needs (Parker, 1996; Stein & Woolley, 1990). Families, therefore, may suffer from a lack of spontaneity in their lives. They may be unable to do anything on the spur of the moment. Plans may be interrupted because of the child's changing needs. When Parker interviewed five married couples and two married women, she discovered that parents of children with a rare NLTl called tuberous sclerosis complex regretted this lack of spontaneity in their lives. Further, these parents felt that their lives were incomparable with the lives of other families because of this disruption. Stein and Woolley also noted that the complex medical needs of the ill children took precedence over family routines.

In one of the first studies in pediatric palliative care (Stein et al., 1989), parents (n=25 families) in the first free-standing pediatric hospice in the world, Helen House in England, were found to experience more psychological difficulties, marital stress, financial, and employment problems than would have been expected from normal samples. Researchers used standardized instruments and semi-structured questionnaires in this retrospective study to examine parents' perceptions of the care offered and the impact of chronic and life threatening illnesses. Only parents were included in the study, although they were asked to respond to questions about the ill children and siblings. In 20 families, the child was still alive. Five out of 25 families experienced significant marital problems. A prospective study was also done in the same hospice to examine

parents' perceptions of the care offered and the impact of a child's life threatening illness (Stein & Woolley, 1990). Twenty-one families from Helen House were matched with a control group who were not receiving or intending to receive hospice services. The groups were individually matched for the child's diagnosis, age, length of illness, and social class. At the time of referral to the hospice, hospice families perceived less overall support compared with the control group (Stein & Woolley). Similar difficulties to those found in the previous study (Stein et al.) were experienced in both the hospice group and the control group (Stein & Woolley). Parents also worried about their child's symptoms, the course of the illness, and about caring for the child if their own health failed (Stein & Woolley).

Family members may focus on the ill child to the exclusion of virtually everything else, including their relationships with each other, extended family, and friends (Gravelle, 1997). During their research at Helen House, Stein et al. (1989) and Stein and Woolley (1990) discovered that families of children with a PLTI had no time for social and leisure activities. Parents were unable to do anything as a couple or as a family when the focus was on the ill child. Difficulties such as marital discord, sibling rivalry, inattention to other children, and loss of relationships with extended family and friends often resulted. Marital difficulties have also been reported by other researchers (Thoma, Hockenberry-Eaton, & Kemp, 1993; Whyte, 1992). Some parents in Parker's study (1996) worried about the effect their child's illness would have on their marriage. They had heard about higher divorce rates in families with ill children. Martinson et al. (1994), however, suggested that while illness can push parents apart, it can also strengthen their love and commitment.

The findings of this research on family structure and patterns of interaction provide important insights on the impact of a child's life-threatening illness on family experiences. However, little is known about how changes in family structure and interactions occur over time. Research evidence when a child has an NLTI is particularly limited. Similar findings have been

reported in studies of families with chronically ill children (Diehl et al., 1991). Parents of children with medically complex needs also reported that extended family members were often afraid of the child (Diehl et al.). This fear may help account for the decreased family contact reported by many families.

In some families caring for chronically ill children, parents shared tasks according to their skill and comfort level. In a study of 30 families, it was reported that during difficult periods of caring for chronically ill children the majority of parents (63%) could focus better on the daily care required when they were able to rely on one another (Clements, Copeland, & Loftus, 1990). There were some families (n=9) where the mother assumed responsibility for care of the household as well as for the ill child. While acknowledging that the burden was heavy, one mother said, "They (men) don't have the inner strength that women do....I think that a woman is a lot stronger than a man" (p.159). Another described her feelings of resignation about her husband's lack of participation in the child's physical care, "He has never participated in the treatments or anything like that. I have asked him, but he said that he didn't know how to do it....knowing his character and everything, he probably wouldn't be able to do a good job of it anyway" (p.159). Single parents (n=2) found caring for the child to be an overwhelming burden. Families drew on many supports to help them. External supports included the health care team, grandparents, friends, informal and formal support groups, and the church. Inner strength came from religion and prayer. Forty percent of the parents reported receiving significant support from their ill child.

Although limited, there is some evidence that families may be able to reduce the impact of their child's illness by reinforcing the family structure (Lesar & Maldonado, 1996; Patterson, Jernell, Leonard, & Titus, 1994; Ray & Ritchie, 1993). In a study of 48 parents of children with HIV infection (n=31) and uninfected perinatal HIV exposure (n=17), Lesar and Maldonado used standardized measures and semi-structured interviews to assess parental coping behaviours and

their mediating effect on the impact of HIV infection on the family system. Eleven participants were foster or adoptive parents, five were relatives, and the remainder were birthparents. These authors suggested that parents who emphasize doing things together as a family and who strengthen family relationships successfully cope with the impact of HIV infection by facilitating family cohesiveness. Caution should be used when interpreting these results, however, because most of the questionnaires were completed by only one parent, usually the mother. Patterson et al. (1994) arrived at a similar conclusion in their study of 48 medically fragile children and their families at home. They determined the behaviours of home care providers that contributed to positive and negative relationships with families. In addition, they identified the child, parent, and community factors that were associated with strain in the parent-professional relationship. Parents completed a number of quantitative instruments, as well as open-ended questions about their experiences and perceptions of caring for their medically fragile child. Family cohesion, family organization, and support from the community reduced the amount of strain parents reported having with home care providers. Whyte (1992) also reported that caring for a child with cystic fibrosis affected the interaction patterns and the coping responses of families. Where there was synchrony between the partners, family functioning remained strong. Whyte found that families needed to move through the transition of seeing themselves as a "normal" family to accepting that they were a family with a health problem. Otherwise, the relationship became very strained.

The findings from research with families of chronically ill children provide important suggestions about the potential impact of a child's life-threatening illness on family structure and patterns of interaction. However, little is known about the transferability of this research to families where a child has an NLTI.

Emotional Impact

The emotional impact on families of caring for a child with a life-threatening illness, including cancer, cystic fibrosis, HIV/AIDS, and NLTIs, is beginning to be documented (Bluebond-Langner, 1996; Clarke-Steffen, 1997; Davies, 1996; Gravelle, 1997; Parker, 1996; Stein et al., 1989; Stein & Woolley, 1990; Wiener, Theut, Steinberg, Riekert, & Pizzo, 1994). Most parents are anxious and worried (Parker; Stein et al.; Whyte, 1992). Wiener et al. examined four dimensions of psychological functioning in 101 parents of HIV-infected children. Scores were generally high on both state and trait anxiety. In addition, almost 25% of the parents were moderately to severely depressed, and another 15% were mildly depressed. Other parents have been found to suffer anxiety, insomnia, and social dysfunction as they struggled to balance the demands of their ill child with the management of everyday living (Stein & Woolley). These parents started or increased their use of tranquilizers, anti-depressants, or cigarettes as they tried to relieve their anxiety. Parents also worried about their child's symptoms, the course of the illness, and about caring for the child if their own health failed (Stein & Woolley). Fear, anger, and feelings of sorrow and loss are also common (Gravelle; Parker; Whyte). In a quantitative study of fathers whose child was chronically ill ($n=23$), one of the most common stressors was worrying about the child's health (Cayse, 1994). Cayse also found that fathers wondered about what the child's future would likely be, and were concerned for their wives' health.

Because some of the PLTIs are genetic, some parents may feel responsible for causing the child's illness. The guilt and anguish that may arise from this sense of responsibility has been noted in research with families where a child has an NLTI (Davies, 1996; Hunt & Burne, 1995; Parker, 1996) or cystic fibrosis (Bluebond-Langner, 1996; Whyte, 1992), and in HIV-infected parents who infected their child (Wiener et al., 1994). Many of the NLTIs are inherited through maternal genes, so mothers may be more likely than fathers to describe a sense of guilt. It may be important to identify which parent feels guilty so appropriate interventions can be implemented.

The tendency of researchers to report data as family-level findings when they have been obtained from a single member of the family may obscure this important element. For example, Davies mailed an investigator-developed questionnaire to 15 families of children with NLTIs. She asked families to describe the factors that influenced how they coped with their child; the strategies they used to cope and how these had changed over time; and what supportive resources they had been told about or used. Unfortunately, although Davies states that families completed the questionnaire, there is no indication of who actually filled out the questionnaire. It is unclear if the results reflect the perceptions of only one member of a family, either the mother or the father, or the family as a unit. One cannot determine exactly who reported feelings of guilt.

There is some evidence that ill children themselves may also experience emotional and behavioural problems, such as anxiety and unhappiness, that, in turn, may further add to the emotional burden parents carry (Stein & Woolley, 1990). Some siblings may also experience anger, sadness, ambivalence towards the ill child, and reduced attention from parents (Ashby et al., 1991; B. Davies & Martinson, 1989; Sourkes, 1980; Stein et al., 1989; Whyte, 1992). In fact, in some families, researchers have reported that siblings may have more problems than the dying child (Spinetta, 1981; Stein & Woolley). Some siblings may worry excessively about their own health, and may be afraid of becoming ill like their brother or sister. Siblings may also have emotional and behavioural problems such as difficulty in dealing with their school work, difficulties in establishing and maintaining relationships with peers, and increased aggression or withdrawal (Gallo, Breitmayer, Knafl, & Zoeller, 1993; Stein et al.). Other authors have also reported nonspecific psychological problems in siblings (Burne et al., 1984; Stein & Woolley). However, mothers in one study recognized that siblings who were well adjusted were minimally affected by the illness (Gallo et al., 1993). Furthermore, several researchers have suggested that living with a chronically ill child may also promote emotional development in siblings. For example, an increase in personal maturity and moral values has been noted in some siblings

(Walker, 1993). The inconsistency of the literature related to siblings of children with chronic physical and cognitive disabilities appears not to have changed since Faux's (1993) review. Faux suggested that siblings may be at increased risk depending on the interactions of such variables as age, birth order, and gender.

Physical Impact

A few authors remarked, almost in passing, that parents of children with PLTIs were exhausted (Gravelle, 1997; Martinson, 1993; Stein & Woolley, 1990). In addition, exhaustion has been observed in mothers of chronically ill children (Stewart, Ritchie, McGrath, Thompson, & Bruce, 1994) and has been recognized as having a significant impact on their lives (Gravelle), although this has not been well described. Clearly, this is an area that requires further exploration. Only one study provides some insight into the possible physical impacts of caring for a child with an NLTi (Leonard, Johnson, & Brust, 1993). The health of mothers of children with disabilities was reported to decline. These parents often encountered such difficulty in managing that they recognized the need for more help. In particular, lifting and positioning the child was reported as physically exhausting for many mothers. More research is clearly needed to understand the physical impact of caring for children with NLTIs.

Financial Impact

Families face many financial costs that are invisible to others, and yet may be a drain on family resources. Sometimes, parents, especially fathers, have been reported to feel they have to work overtime to ease their financial burden (Clarke-Steffen, 1997). Often, particularly in a country such as Canada where government health plans ensure a basic level of coverage, people assume that all costs are covered by insurance. Most insurance plans, however, do not pay for incidental costs. The cumulative effect of these costs may take a toll on families.

Some researchers in pediatric palliative care have identified high cumulative, monetary costs related to buying medications, special diets and equipment, or frequent attendance at health

care facilities with the associated costs of travel, food, and telephone calls to the family left at home (Birenbaum & Clarke-Steffen, 1992; Stein et al., 1989; Stein & Woolley, 1990). In addition, they have reported indirect costs, such as mothers reducing their work hours or giving up their jobs to take care of the child, that result in a loss of family income at the same time that financial costs are increasing (Parker, 1996). Some fathers refused promotions or transfers, while others became unemployed or bankrupt (Birenbaum & Clarke-Steffen; Stein et al.; Stein & Woolley). In addition, those parents with insurance may be unable to change jobs because new insurance companies may not cover them. Even when families have insurance coverage, either by private coverage as some do in the USA or through government health plans as in Canada, families may still incur great financial costs when a child is ill.

Schweitzer, Mitchell, Landsverk, and Laparan (1993) noted that, though families usually want to keep their child at home, the cost of nonreimbursed expenses may be enough to exclude many families from participation in a home-based program. These researchers used two sources of data to estimate direct costs (such as wages of health care workers, medication, and special clothing and supplies that would not have been purchased otherwise) and indirect costs (including lost wages where a caregiver lost a job, gave up leisure time, or took vacation, and volunteer care by friends, neighbours, and other volunteers). Retrospective data on provider utilization and duration in an American home-based pediatric hospice were collected for 177 families. Twenty-seven of these families provided prospective data through telephone interviews and expenditure diaries. Weekly incidental expenses averaged \$42, and average indirect expenses amounted to \$140 per week. The total average cost profile for home care episodes was \$4,808. Direct personnel costs averaged \$2,850, direct incidental costs were \$446, and indirect costs amounted to \$1,478.

For many families of chronically ill children, a high percentage of weekly income may be spent on out-of-pocket expenses. Studies have consistently shown that these expenses may be a

significant drain on family resources (Diehl et al., 1991; Ray & Ritchie, 1993; Walker, Epstein, Taylor, Crocker, & Tuttle, 1989; Youngblut, Brennan, & Swegart, 1994). Out-of-pocket expenses reported in one study of 910 parents included travel expenses, parking fees, and child care for other children, as well as other expenses (Walker et al., 1989). Diehl et al. (1991) also reported a lack of financial assistance to middle income people and difficulty in finding out what assistance might be available.

Spiritual Impact

The impact of caring for a chronically ill child on experiences related to spirituality and faith have been described by a few researchers. There is evidence that many parents seek out spiritual guidance and support (Davies, 1996). Spirituality and faith have provided both emotional and network support for many parents. Faith has been shown to be a key factor in a family's ability to keep an ill child at home (Davies, 1996) and in promoting positive adaptation among families of children with disabilities (Bennett, DeLuca, & Allen, 1996; Youngblut et al., 1994). Bennett focused on positive adaptation and investigated the informal resources that parents use across the family life cycle. Interviews with 12 parents of children with disabilities ranging in age from 15 months to 30 years revealed that religious beliefs were particularly supportive. Faith in a higher power has been associated with higher levels of coping and hope in family caregivers of ill adults (Herth, 1989; Herth, 1993; Kaye & Robinson, 1994). It has also been reported as an effective coping strategy in a number of different illness situations (Cayse, 1994; Lesar & Maldonado, 1996; Schwab, 1990; Steele & Fitch, 1996). Some families of chronically ill children report receiving inner strength from religion and prayer (Clements et al., 1990; Rivera, Stoner, & Groothuis, 1996). A strong belief in God may provide caregivers with the strength and support they need to continue caregiving.

Summary

It is not unrealistic to expect that families may undergo many changes when a child has a terminal illness. Normal patterns of interaction may be disrupted, and families may need to learn to adapt to new challenges and changes. There is evidence to suggest that family structure may be at risk of deteriorating as energy, time, and resources are directed towards the ill child. Living with a child who has a PLTI may also cause emotional, physical, financial, and spiritual disruptions. However, it appears that some families may be able to cope with changes better than other families. For example, families where parents share tasks and rely on one another may have less difficulty in dealing with the changes and with the care of the ill child. Clearly, a better understanding of the impact of caring for a child with an NLTI is needed. The situation of caring for such a child presents some unique challenges that we are only beginning to understand. We need to know more about these impacts and challenges to learn more effective ways of supporting these families.

Factors Influencing Families' Experiences

Families' experiences do not occur in isolation. Instead, they are influenced by a number of factors that may be internal or external to the family. Some factors that are thought to influence the experience include the location of care, communication among family members and between families and professionals, and level of family functioning. These factors will be examined to provide a context for understanding families' experiences.

Location of Care

The physical environment in which care occurs may affect the family. While there are no pediatric studies that have asked dying children their preferences about location of care, it is believed that most children would prefer to be at home (Kopecky et al., 1997). The importance to families of the location of care is demonstrated in studies involving three different programs

providing home-based palliative care for children with terminal illnesses (Duffy et al., 1990; Lauer & Camitta, 1980; Martinson, 1993; Martinson et al., 1986).

Duffy et al. (1990) surveyed eight parents of children with central nervous system tumours who had died while receiving home-based palliative care. Their overall satisfaction with the program was high compared to the care they had received prior to admission to the program. The component of the program that was most satisfactory was being able to care for the child at home with the support of professionals. In another study (Lauer & Camitta, 1980), families of children with cancer who received home care (n=42) reported that they wanted the child at home. In addition, the medical and nonmedical financial burdens of inpatient and outpatient hospital care were reduced when the child died at home.

The Home Care for the Child with Cancer Project is a comprehensive study that was conducted from 1976 to 1980 in three large American institutions (Martinson, 1993; Martinson et al., 1986). The purpose of this project was to evaluate the feasibility of home hospice care for families. Primary care was provided directly by families with strong nursing support. Nurses made home visits and were available on call 24-hours-a-day, 7-days-a-week. Fifty-eight of the 64 children referred to the project over the reported 2-year study period died; 46 of them at home, 11 in hospital, and 1 en route to hospital. The children had a wide range of cancer diagnoses. Their parents participated in semi-structured interviews one month post-death to review the process of home care, parental assessment of care, and financial aspects of home care. Martinson (1993) reported that parents whose child died at home had fewer negative and more positive comments about their child's home care, compared with those whose child had died in hospital. Parents noted that fulfilling their child's wish to get home had positive effects. They perceived the child to be happier, more secure, and relaxed once home and no longer separated from family and friends. Children were also able to live more normally and have more control over their lives. The illness was very disruptive for parents, but they believed that home care

reduced this disruption and allowed them to have more control. Parents also enjoyed being able to care for their own child. They valued being constantly nearby. Negative aspects of home care included stress, exhaustion, difficulty in keeping well-meaning visitors away, and the need for more follow-up and management of medication.

Invasion of family privacy has been identified as an important issue for some families caring for a child at home (Leonard, Brust, & Patterson, 1991; Patterson et al., 1994). Families are often dependent on home care professionals. Yet, the regular presence of others in the home may increase the strain on families. Two major themes that emerged from analysis of open-ended questions in the Patterson et al. study were related to home care staff's competence, and attitudes of nurses towards the child, parents, and the rest of the family. The greatest difficulties in parent-professional relationships were experienced by mothers when they received more help from home health aides, as opposed to professional nurses or home helpers. One of the challenges noted by this study was the need for professionals to balance supporting the family in a non-intrusive manner while providing sufficient support.

Findings from these studies must be interpreted cautiously because the studies were retrospective, and from the perspective of parents, usually the mothers. Consequently, the results may be biased by the recollections and experiences of only one member of the family. They may not reflect the family's experience. Despite the limitations of the previous research studies and the potential drawbacks of home care, results suggest that many families want to care for their ill child at home wherever possible. Finding ways to support families requires an in-depth understanding of families' experiences and how they manage at home. Yet, there is a paucity of research that examines families' experiences at home when one of their children has a PLTI. Although it appears that some families want to have their child at home, we only have a cursory understanding of the impact on family life of caring for a child with a PLTI.

Communication

Although open and effective communication may be important for families as they care for their ill child (Clarke-Steffen, 1997), problems related to communication have been identified among family members (Clarke-Steffen; Whyte, Baggaley, & Rutter, 1997), and between families and health care professionals (James & Johnson, 1997). Issues related to communication have been addressed by researchers in the context of families' experiences. Sharing information with the ill child and siblings can be especially difficult for some parents (Clarke-Steffen). Parents often struggle to find a balance between protecting children by withholding information and providing sufficient information to maintain a trusting relationship. Sometimes, parents may have discrepant views of the situation. Even when they try to address their differences, these issues are often a source of ongoing conflict. A qualitative study of 16 families where a child had congenital heart disease, asthma, diabetes mellitus, or cystic fibrosis revealed similar findings (Whyte et al.). The investigators reported that most families had difficulty maintaining good communication, even when parents had strong communication skills prior to the child's illness. This block in communication between parents contributed to strain in marriages and affected parents' ability to continue caring for their child. Other researchers have also noted that the spousal relationship in similar contexts has an important influence on parents' mental health (Deatrick & Knafl, 1990; Nagy & Ungerer, 1990; Ray & Ritchie, 1993).

In some cases, communication about the child's condition may be unclear between health care professionals, such as when a child's status changes from active treatment to palliative care (B. Davies & Steele, 1996). Researchers have noted that this lack of communication may create gaps in the support services provided to patients and families (Davies, 1996; Singleton, 1992). Moreover, the bureaucracy involved in obtaining services can be very frustrating for parents (Gravelle, 1997). Faulty communication may also cause anxiety in parents that impedes satisfactory care. In her study of families where a child had an NLT, Davies reported that

families were often followed by multiple services within the hospital. Having one person, who knew them and their child, co-ordinate their care, facilitated their connection to the hospital and made it easier for families. Until that link was in place, they felt isolated, alone, and unsure of their ability to care for their child at home. Other researchers have also identified the need for a competent case manager to ensure adequate communication (Diehl et al., 1991; Gravelle; Whyte, 1992; Woolley, Stein, Forrest, & Baum, 1991). In the U.K, the term "cornerstone carer" was used by Woolley et al. during their research at Helen House (Stein & Woolley, 1990). These researchers defined a "cornerstone carer" as the person who provided central support or who enlisted or co-ordinated appropriate help. Eleven out of 45 families in this study did not have a "cornerstone carer." Of those 11 families, 9 had children with NLTIs. Families where a child has an NLTI may, therefore, be particularly vulnerable to communication difficulties.

Families appear to have a variety of experiences in communicating with health care professionals. In a retrospective, qualitative study of 12 parents of 8 children who had died from cancer, parents reported feeling cared for when professionals spoke with them and treated them with respect, compassion, and sensitivity (James & Johnson, 1997). Conversely, a professional's failure to listen to parental concerns caused frustration for parents. Moreover, parents felt abandoned by caregivers who physically and emotionally withdrew from them. Parents in Parker's study (1996) also reported feelings of frustration, anger, and hopelessness when health care professionals refused to take parents' concerns seriously.

Researchers have determined that it may not be easy for families to communicate with health professionals (Clarke-Steffen, 1997; Diehl et al., 1991). For example, Diehl et al. used focus group interviews to identify parents' perceptions of their needs when a child has medically complex needs. These parents identified the health care system as a major source of distress and frustration because of its fragmentation of care. In addition, parents reported a feeling of "them against us." They felt that professionals, especially doctors, did not listen to parents or give them

the respect they deserve. Parents thought they needed to be assertive and informed because professionals seemed uncomfortable with the child, did not spend enough time with the parents, and blamed the parents for the child's behaviour even though the child might be sick.

Professionals who did a good job were the exception.

Parents' need for honest and appropriate information so they can manage their situation has been well documented in studies, and includes those families who have children with cancer (Clarke-Steffen, 1997; James & Johnson, 1997), cystic fibrosis (Bluebond-Langner, 1996), NLTIs (Davies, 1996), and other chronic or life-threatening illnesses (Diehl et al., 1991; Gravelle, 1997; Walker et al., 1989). Information may be empowering (Clarke-Steffen; Gibson, 1995; Gravelle), and allow families some control in a situation where they have little control. It may also help to reduce uncertainty (Cohen, 1995; Galloway & Graydon, 1996; Selder, 1989). Yet, some researchers have shown that information needs are not always met (Clarke-Steffen; Walker et al., 1989). In a mailed survey with a convenience sample of parents whose children had various chronic illnesses (n=910, 38% return), communication and information needs were ranked as important, though frequently not met (Walker et al.). Many parents wanted more communication with health professionals. They needed to obtain information about programs and services, financial planning for the future, the child's particular health problem, financial aid and insurance plans, and the treatment prescribed. Barriers faced by families when looking for information may include complex medical language, reluctance of some professionals to disclose information, and negative attitudes of those providing the information. Moreover, in situations where there is little information about a disease entity, as is the case with NLTIs, the task of providing the information parents need becomes more difficult. Thus, even when professionals are willing to share what they know, there may be no information available. In such cases, the scarcity of information has been seen to contribute to increased uncertainty and frustration in these families (Davies; Parker, 1996).

Problems in communication have also been attributed to discrepancies that arise when parents need to talk about their feelings related to the child's death and dying and family members and friends are unwilling to discuss these issues (Diehl et al., 1991). Our understanding of the complexities underlying communication problems for families has been extended by Jefidoff and Gasner (1993) in their study of Israeli parents of a dying child. They found that even when families can discuss their fears and talk about the child's imminent death, they may be unable to articulate the more unconscious and complex needs, such as those related to understanding changes in family dynamics that may occur in the final stages of the disease or describing feelings about the funeral and burial, unless they have the assistance of skilled hospice staff. The investigators suggest that families' ability to recognize and deal with these potential stressors puts them at higher risk for further problems. The findings from this study (Jefidoff & Gasner) must be interpreted cautiously however, because the researchers did not say how many families or parents were included in the study. Moreover, the results from this Israeli study may not be generalizable to other cultures.

In summary, issues related to communication appear to play a significant role in shaping families' experiences when they are caring for an ill child. Our understanding, however, remains limited because of the paucity of research that has addressed these issues. Further, the complexity of the dynamics involved in communication needs to be explored in order to learn how to provide effective care to families of children with NLTIs.

Levels of Family Functioning

The concept of family functioning has received considerable attention from family theorists, family therapists, and others who focus on family care (Friedman, 1998; McCubbin & McCubbin, 1989; McCubbin & McCubbin, 1993; Olson, Russell, & Sprenkle, 1983), but only minimal attention in palliative care. It is not improbable that a family's ability to adapt to the changes imposed by the child's illness and impending death may depend in a large part on their

level of functioning. One group of researchers (B. Davies, Spinetta, Martinson, McClowry, & Kulencamp, 1986), however, have made an important contribution to our understanding of family functioning in the context of pediatric palliative care. In their study of 111 families whose child had died from cancer, the investigators conducted semi-structured interviews and rated family functioning using a five-point scale. The global rating of each family's overall level of functioning was derived from the researchers' judgements based on clinical assessment of the family during the interview, the family's responses to the interview questions, and how the family compared to other families. The dimensions of functioning were viewed as occurring along a continuum of functionality and were not judged as good or bad.

Families at the extreme ends of functionality were compared and contrasted. The more functional families freely discussed the dead child, the illness and death context, and the family's responses since the death. They shared views with other members of the family and were able to disagree without conflict. More functional families recognized that others were also grieving, and that differences in grieving were alright. They were empathetic and supportive. On the other hand, some families did not have freedom of expression and one member frequently spoke on behalf of another. Members of these less functional families focused on their individual grief and did not empathize with others, or even recognize others' grieving or differences in grieving. Functional families were aware of a process occurring. They could describe their feelings about events, such as the funeral, rather than only being able to concentrate on the content, such as the location of and attendance at the funeral. They anticipated special events, such as Christmas, as being difficult and tried to prepare constructively. They could also see how they had changed. Less functional families, on the other hand, made no plans to cope with difficult times, did not share their feelings, and did not seem to change. They seemed resigned to everything as if they could not change it (B. Davies et al., 1986).

Functional families also acknowledged their grief and questioned how they could cope with the sadness of their child's death, while seeing that something positive had come out of the experience. Returning to a normal life which included frivolity was important to functioning families, and they integrated their experience into their life. They wondered if they could or should have done something different to help their child. But, they were not regretful. They saw these feelings as part of the mourning process. They continually changed their focus of hope as the child's condition deteriorated. Thus, although they still hoped for a miracle, they were realistic about the prognosis. These families remembered the good and the bad things about their child. In contrast, other less functional families suppressed their natural feelings and often made comments related to religion that reflected a belief that they could not question events because it would show a lack of faith. Some had difficulty in accepting that their child was dying. They often fought the diagnosis till the end without acknowledging the inevitability of death. They often idealized their dead child and seemed to forget that he or she had once been a normal child who misbehaved on occasion. Less functional families regretted what they had done or not done that may have contributed to their child's death. Their lives were based on their experience of having a child die. Everything else was seen as frivolous compared to that experience.

Functional families were flexible in their roles (B. Davies et al., 1986). They acknowledged that one child was no longer there and did not expect another child to fill the gap. Less functional families were often rigid about roles and expected another child to take over the roles of the dead child. Some parents imposed the roles, some family members self-imposed them, and some parents expected surviving children to assume the characteristics of the dead child. Gender-related role rigidity was more obvious in less functional families in which the father would not express grief over the dead child. More functional families were able to ask for and use help from a variety of sources. They could also turn down unwanted help graciously. Other families were uncomfortable or lacked confidence to ask for help and felt very alone.

Functional families recognized their loss and the feelings of sadness, anger, and depression that accompanied it. But, they were able to reorganize their life and move on. They knew that they were not and could not be the same as they were before their child died. Others did not want to reorganize and tried to keep everything as it had been. They remained sad, depressed, or angry and could not move on.

Functional families accepted that they were vulnerable and admitted the pain they suffered. They always tried, though, to see the good things in life as well. Lower functioning families defended their feelings of vulnerability. They could not give themselves permission to laugh because they believed that the enormity of their loss prevented them from ever having fun again. Functional families also reassessed their lives in view of their experience and questioned their previously held beliefs. Quality of life and person-related values often became more important to these families. Although they accepted their child's death as fate, they also vented their anger at God. They were able to continue to believe in God even though they did not like what had happened to them. Other families accepted the hand of fate without question and saw God as all good or all bad. They either never questioned why or they left their religion.

Expression of sadness and tears was accepted by functional families as part of the grief process. Conversely, dysfunctional families often equated sadness with craziness. Feelings might come out too strongly at other times of stress though, such as when a family pet died. Functional families had low control over their grief. They discussed their feelings and shared their sadness. They were sensitive to others. Dysfunctional families had a high control of their grief. Yet, nonverbal behaviour was often an indicator of underlying emotions. The more functional families were open, flexible, and adaptive. They could drop old friends who did not share their new perspectives and they made new friends who did. They could move on with their lives. They often became reinolved with children with cancer to help others benefit from their experience. Less functional families were bitter about old friends who could not cope with the child's death.

They did not want to make new friends. If they became involved in healthcare issues, it was only to help themselves work through their difficulties (B. Davies et al., 1986).

In another study (B. Davies et al., 1995), family level of functioning was also related to how families managed the transition of fading away when one of their adult members was dying with advanced cancer. In this three phase study, the researchers used grounded theory methodology to understand how families experience their lives when one of their members is dying. Twenty-three families in total were interviewed. Participation came from 66 family members, including the terminally ill adults. Again, dimensions of functionality occurred along a continuum.

Some families used past experiences as opportunities for growth and learning, while others dwelled on the negative aspects (B. Davies et al., 1995). In some families, members described a broad range of feelings and acknowledged vulnerabilities and paradoxical feelings. Others avoided talking about the impending death and were often angry. Some families identified and discussed problems through mutual support and togetherness. They resolved the problem by taking control of the situation. These families learned from the problem and its solution. Other families concentrated on the emotional issues without trying to find a solution. They were often bitter and blamed others for the situation. As a group, they showed consensus. But in individual interviews, they often blamed other members of the family for the problem. Families who used multiple resources and support were usually satisfied with the result of the help. Some other families were reluctant to seek or accept help and were often dissatisfied with the help they received.

Some families recognized each member for their valuable contributions to the patient's well-being and concentrated on keeping the patient well cared for without seeking attention for themselves. In contrast, in other families, each individual seemed to focus on their own needs and not recognize the contributions or needs of others. Flexibility in allocating responsibilities

usually resulted in families who were happy with their decisions. Conversely, families who tried to hold on to old roles could not adapt easily. Members sometimes felt obligated to provide care, rather than wanting to do it. Some families tolerated differences inside and outside the family, even though they did not necessarily agree with a particular view. Where families were in agreement about the family identity, they could provide a supportive environment for their members. Some families did not have a family identity. Instead, they only had personal characteristics. The "truth" was often different in the group compared with individual interviews. There was often a history of abuse in these families. These families often had difficulty in tolerating differences outside the family as well (B. Davies et al., 1995).

Clearly, there are differences in how families deal with situations. How these differences affect provision of care is not as clear. Negotiation and mediation may be critical when working with families who are not cohesive. Further research is necessary to understand how the dimensions of functioning affect how and when services, options, and resources should be offered. This understanding is necessary to develop the range of interventions that may be required by families with varying degrees of functionality.

Conclusion

There is a dearth of research and knowledge to guide practitioners in providing optimal care for families where a child has a PLTI, particularly a neurodegenerative disease. While the available research provides an important beginning, the findings must be viewed as tentative because of small sample sizes and the scarcity of studies. In addition, the retrospective nature of some of the studies may have resulted in important information being forgotten or under-reported. Knowledge about families where a child has a PLTI is steadily increasing, but very little research has focused on families of children with NLTIs. While researchers investigating children with chronic illnesses provide some support for these findings, one must be careful of

drawing firm conclusions. Any differences between families of children with NLTIs and those with chronic diseases may not be apparent in the limited research that is available.

The research to date provides a cursory understanding of the impacts of families' experiences when a child has a NLTI, and the factors that influence those experiences. Research indicates that all members of the family may be adversely affected by a child's life-threatening illness. Most of the available research, however, has been from the perspective of the parents, and sometimes only the mother. There have been few attempts to capture the views of the ill child and his or her siblings. Yet, relying on adults to identify impacts on children, whether ill children or siblings, may neglect the children's fears and anxieties and may fail to capture their experiences. It is critical that all members of a family, including the ill child, are asked about their experiences.

A number of the studies with families of children with a PLTI used quantitative methods to obtain data (Lesar & Maldonado, 1996; Martinson et al., 1986; Nagy & Ungerer, 1990; Stein et al., 1989; Stein & Woolley, 1990; Thoma et al., 1993; Wiener et al., 1994). Some of these instruments were developed for the studies and underwent little psychometric testing, so their reliability and validity are unknown. Others were standardized psychosocial instruments that have limitations when completed by such families. Many psychosocial scales were developed and normed on college students or psychiatric patients. They may, therefore, have little relevance to families where a child has a PLTI. In addition, most instruments designed for healthy families or families of chronically ill children have not been subjected to validity and reliability testing in families where a child is dying. Thus, limited confidence can be placed in the results obtained from such measures. Finally, pre-existing instruments can only capture discrete parts of an experience. Families would need to complete many more instruments than were used in these studies in order to fully describe their experience.

The focus on rich, detailed descriptions in some of the qualitative studies has uncovered important aspects of families' experiences when a child has a life-threatening illness.

Unfortunately, the studies of families where a child has an NLTI are very limited in their descriptions. There is a need for more in-depth studies to uncover the experiences of families as they live with a child who has an NLTI. Moreover, it is important that all members of a family have the opportunity to contribute their perspective. Retrospective studies that are done after a child has died may fail to reveal the depth and breadth of families' experiences. Instead, research is needed during the experience itself. Qualitative research methods may be the most effective way of capturing the full richness of families' experiences when a child has an NLTI.

Research Questions

Specific research questions that guided this study were:

- 1) What is the family's perception and experience of living with a child who has a neurodegenerative, life-threatening illness?
- 2) How does the family's perception and experience change over time?
- 3) What is the impact on the family of living with a child who has a neurodegenerative, life-threatening illness?
- 4) What factors do families perceive enhance or undermine the family's ability to care for the child with a neurodegenerative, life-threatening illness?

Definition of Terms

For the purposes of this study, some terms were defined as follows:

Diagnosis Period: The beginning of living with a child's neurodegenerative, life-threatening illness; when a family first learns about the child's illness. This period of time lasted from initial diagnosis until one month after diagnosis.

Family: A complex unit that is composed of individual members, but that has distinct attributes of its own. Ideally, the family is self-defined and includes whoever the family says it includes.

The family unit in this study was limited to those members with whom health care professionals most often interact and who usually play a key role in pediatric palliative care - the ill child, parents, and siblings.

Ill Child: A child, from infancy to 17 years of age, who had been diagnosed with a neurodegenerative, life-threatening illness.

Neurodegenerative Life-Threatening Illness: Any disease that led to progressive destruction of the nervous system and was expected to cause premature death in childhood or early adulthood.

Pediatric Palliative Care: Active care to children and their families when cure is no longer possible. Alleviation of symptoms, psychological, spiritual, and social problems is paramount. The goal of pediatric palliative care is to assist children and their families to achieve the greatest possible quality of life. Pediatric palliative care encompasses respite, terminal, and bereavement care.

Respite Care: Support provided to families of a child with a neurodegenerative, life-threatening illness that relieved them of the continuous 24-hour care of the child for a period of time. Respite care is provided in and out of the child's home.

Respite Period: The time from one month after diagnosis until one month prior to the expected death of the child during which the family lives with and manages the child's illness.

Terminal Period: The end of living with a child's neurodegenerative, life-threatening illness; when the child was expected to die within one month.

Chapter IV: Research Design and Implementation

Theory is critical to the development of any practice discipline such as nursing. The generation of knowledge for practice provides evidence on which to base interventions. The focus of this study was on generating a detailed, contextually-grounded description and theoretical explanation of the experiences of families with a child who has a neurodegenerative, life-threatening illness (NLTI). The choice of research design was made on the premise that the research design in any study must be suitable for the state of existing knowledge about the questions being asked (Siegel, 1983). Little is known about family experiences when a child has an NLTI. In addition, families themselves are the most pertinent source of information about their experiences. Qualitative methods are most appropriate when asking questions dealing with subjective experience and perceptions with situational meaning. Grounded theory is a qualitative research method that is particularly suited to family research because of its focus on social processes (Daly, 1992). Therefore, grounded theory had the potential to develop theory about family experiences. Consequently, the primary analytical method used in this study followed the procedures of grounded theory (Glaser & Strauss, 1967; Strauss & Corbin, 1990).

Symbolic Interactionism

The design of a grounded theory study is guided by the underlying symbolic interactionist theory in which shared meaning creates human behaviour. Symbolic interactionism is a theoretical perspective that is often used to guide research and practice in the family field (Klein & White, 1996). Symbolic interactionism characterized the theoretical perspectives of scholars such as pragmatists including Mead, James, Dewey, and Pierce, and sociologists including Thomas, Park, Cooley, and Wirth (Lindesmith, Strauss, & Denzin, 1975). In the early 1930s, Mead (1934) delineated an ontological position that was later articulated as symbolic interactionism by Blumer (1969). The symbolic interaction school of thought relies heavily on the pragmatic philosophical tradition which has its roots in the belief that a phenomenon only has

meaning if it can be directly or indirectly applied to a specific situation. In addition, it argues that an individual is in constant interaction with the environment and chooses which stimuli to respond to (Burr, Leigh, Day, & Constantine, 1979). Interactionists believe that understanding of human behaviour requires learning about the beliefs and values that individuals get from interacting with others. The best way to understand the behaviour of humans is to understand the meanings and values that occur in people's minds and that cause behaviour. However, these mental variables are affected by antecedent variables, such as feedback from intimate interactions; perceptions of generalized conditions in the social situation; and contextual variables like social status or social norms. In addition, the physical self is different from the social self. The "I" part refers to the unpredictable and spontaneous parts that make a person unique, whereas the "Me" part is determined by learned social roles. Because the "I" part of the self will influence what goes on in the mind, there is always an unpredictable component to the mentalistic variables (Burr et al.).

Unlike the approach of many social scientists, symbolic interactionists do not assume that culture is composed of commonly shared signs and symbols that are uniformly understood. Symbolic interactionists believe that one cannot understand a person's behaviour unless one understands the meaning of an event from the individual's perspective. Thus, cultural meanings and social behaviour are inexorably entwined. How a situation is interpreted is based on the meaning the situation holds for the individual. Meaning arises from a number of variables including past experiences, language, educational level, ability, and cultural background (Klein & White, 1996). Symbols are used to constitute meaning, and meaning is negotiated through symbols. Humans decide how to behave on the basis of learned symbols and their beliefs about the importance of meanings. Therefore, individuals act according to shared meaning, and behaviour is influenced by the meaning of the ideas in the mind.

Guided by symbolic interactionism, a researcher needs to explore the meaning given to situations by the people involved. One cannot assume that meaning is shared by all of the participants. A researcher must try to understand the world from the participant's perspective. Thus, objective measures are not sufficient to obtain information. Instead, formal and informal interviews with participants are required. In addition, one cannot observe the part of the self that is "I," only the part that is socially defined as "Me." Consequently, one must again obtain subjective information by discovering the person's perspective. Informal and formal interviews were, thus, an integral part of understanding and studying families in this current research study.

Since meaning is constructed through interaction and meaning creates behaviour, then behaviour must be studied at the interactional and at the symbolic level. In order to understand human behaviour, a researcher must examine interactions in context. Therefore, behaviour needs to be observed in natural settings. Participant observation can be used to look for interaction, patterns of interaction, and their consequences. In addition, analysis of the interactions must include the symbolic level, so informal questioning of participants must be done immediately after an interaction to clarify the participants' self-definitions and shared meanings. A researcher cannot assume the meaning of an action or interaction, but must check out her assumptions and seek information directly from the participants. In studying families in which a child has an NLTI, participant observation was important to provide data about the process of living with such a child, and about how families managed caring for their ill child.

Setting

Data collection took place in the homes of families whose child had an NLTI. The purpose of this research was to understand how families managed their lives with such a child, as well as how their lives were affected by the illness. The researcher offered to accompany families during their daily lives in order to gain this perspective. Families consistently explained

that their lives revolved around the ill child in the home. Interviews and supplementary observations, therefore, all took place where the families suggested - in their homes.

Selection of Participants

Families were recruited for this study through two sites: a pediatric hospice care program and a children's hospital. Both sites were located in the Lower Mainland of British Columbia. Families were eligible for the study during the period of time in the child's illness when they may require respite care. For the purposes of this study, this period was defined as lasting from one month after diagnosis until one month prior to the expected death of the child. Where a child had just recently been diagnosed, families were excluded because it was anticipated that they would lack the depth of experiences that would make them key informants for this study. Those families in which the child had reached the terminal period of the NLTI were also excluded because of the potential stress of this period on families. In addition, it was anticipated that those families may have different needs and experiences from families requiring respite care. For the purposes of this study, family was limited to those members with whom health care professionals most often interact and who usually play a key role in pediatric palliative care - the ill child, parents, and siblings.

Each member of the family who could communicate verbally and or in writing was asked to participate. The ill child was part of the study in whatever capacity was possible, such as being observed during care. All of the ill children were precluded from full participation because of profound mental impairment or an inability to communicate. Families were not excluded from the study because the ill child was unable to participate, nor because one parent or siblings were non-existent, unavailable, or unwilling to participate. This research included those children, parents, and siblings who were willing and able to participate. Initial contact with families was made by the hospice and hospital staff. Families were considered potential participants if they met the following eligibility criteria:

- 1) The ill child had been diagnosed with a neurodegenerative, life-threatening illness. Where an illness is not life-threatening, families may face different issues. Neurodegenerative illnesses may create unique circumstances for families.
- 2) The ill child was 17 years of age or younger. The context of this research was pediatric palliative care.
- 3) Families were living in the period of time in the child's illness when they may require respite care, as defined in this study. At least one month had elapsed since diagnosis and death was not expected by clinical personnel for at least one month following participation. The specific context of this research was the respite component of pediatric palliative care.
- 4) At least one adult, probably a parent, who lived with the child and was involved in the child's care was willing to participate in the study. Single parent families were not excluded.
- 5) Those adult family members who wished to participate were able to read, write, speak, and understand English. Informed consent and participation in interviews required comprehension of written and verbal English.
- 6) Those siblings who wished to participate were able to communicate verbally and or in writing in English. From about age seven years, minor children needed to give their assent in addition to parental consent. Adult siblings were eligible for participation.
- 7) The family was emotionally and physically capable of participating in the study. The researcher and the clinical personnel were to make this judgement. Those families who were considered at risk were not approached.

As data analysis and collection proceeded, theoretical or purposive sampling was used to ensure that the sample was appropriate. Families were deliberately selected for their potential contributions to the developing theory, and according to the theoretical needs and direction of the study. This approach ensured that the evolving theory was representative of the phenomena being investigated, and allowed for examination of the full range of variation in emerging

concepts in order to develop theory. To extend the conditions under which actions and interactions occurred, interviews occurred with families who required varying amounts of respite care. In addition, some children were deteriorating slowly while deterioration was occurring rapidly in others. Data collection continued until the data were saturated, that is until no new data were obtained from interviews and the phenomena were richly described.

The Sample

Twenty-nine participants were interviewed or observed in this study. Of the eight families who participated, two families had two children who were affected by an NLTl. Therefore, 10 sick children in total were observed. Six of the children were boys and the ages ranged from 3-13 years. At the time of the study, the children had been diagnosed for two and a half to six years. None of these children could be interviewed because they were unable to communicate verbally due to the nature of their illnesses. While all the children had neurodegenerative illnesses that would eventually lead to their death, the actual diagnoses will not be named to facilitate confidentiality of families. Most of these illnesses affect very few children throughout the world, perhaps less than 100 for each diagnosis. Indeed, some of these children represent the only known case of a particular illness in British Columbia. Identifying their diagnoses would effectively identify the children and families. While the different illnesses are manifested in many ways, common attributes such as changes in verbal ability, changes in motor skills, and the proliferation of feeding disorders meant that these families experienced many of the same opportunities and challenges as they lived with the illness.

There were only three families in which the ill child had siblings. Usually, parents chose to have no more children once they learned about their child's diagnosis because these NLTIs are often genetic. Out of four siblings, three, all female, had been born before the child was diagnosed. The other sibling was male, and the siblings' ages ranged from 2-9 years.

In the majority of families, the parents were married and lived together, although there were difficulties in most marriages. One couple had been married but was now separated, another couple was also separated but had never been married, and in the eighth family, a single father cared for his child alone. The mother was not involved in that family's life at all. The length of the marriages ranged from 8-15 years. Parents' ages ranged from 28-48 years and most had completed high school, although educational levels varied from grade six to a university degree. Occupations of the parents included those who were currently unemployed, often because they were caring for the ill child, blue and white collar workers, and professionals.

Socio-economic diversity was apparent with annual family incomes ranging from \$11,000 to \$112,000. Four families subsisted on incomes of less than \$15,000 per year, while the other families earned \$50,000 or more. However, all families reported a substantial drop in actual or anticipated income due to the child's illness.

Geographic diversity was restricted to the Lower Mainland of British Columbia, despite efforts to contact families in other parts of the province. However, location of the tertiary children's hospital is important for families, so families often choose to live close to the hospital. While urban and suburban settings were represented, it was somewhat difficult to distinguish what difference geographical location might make. Some families in the suburbs felt they were very close to the city, yet others who lived in the same area stated that the trip to the city was a major event because it was so far away. Of the eight families who participated in this study, all but one family identified with the dominant Caucasian Canadian culture. The other family came from an East Indian background.

Data Collection and Procedures

In accordance with grounded theory methodology, the main method of data collection was in-depth interviews with families supplemented by participant observation. Data collection was facilitated through the pediatric hospice care program and the children's hospital. Initial data

collection occurred over a period of one year. During the following year, the evolving analysis was shared with the families. After the first interviews and observations were completed with all eight families, each family was sent the preliminary analysis by mail. A letter was enclosed inviting families to contact the researcher if they had any comments. The letter also stated that the researcher would call each family (see Appendix 6). One family called immediately on receipt of the package. A second interview took place with this family in the family home. All other families were called by the researcher. Subsequent interviews took place with these families by telephone. Two families requested that they receive major iterations of the evolving theory so they could make comments. The data were sent by mail and these families were interviewed by telephone on a few occasions. The other families simply requested a copy of the completed research report.

Initial contact with potential participants was made by clinical staff, all of whom were nurses. The researcher met with staff to explain the study, to outline the eligibility criteria, and to answer any questions staff had. Ongoing communication with staff was critical in ensuring accrual of participants into the study. Other researchers (B. Davies et al., 1995; McCorkle, Packard, & Landenburger, 1985) have suggested that physicians, someone designated by the physician such as a nurse, or a family member may act as a barrier to recruitment of potential participants. Further, B. Davies et al. suggested that nurses in particular may be reluctant to tell families about a study because they often assume the role of patient and family advocate, and may try to protect the families from a seemingly stressful imposition. Sometimes it may be unclear whether a person is really unsuitable for a study or if the advocate's attitudes and prejudices have influenced the decision (Ling & Penn, 1995). Therefore, to facilitate accrual, the researcher attempted to have frequent face-to-face communication with the professionals involved in recruitment and to maintain a high profile. However, many of the contacts actually occurred by telephone and electronic communications. Despite the researcher's best intentions

and attempts to encourage staff participation in research by discussing why and how research is done, by pointing out the benefits of research, and by highlighting the ways that patients' and families' rights are safeguarded (Raudonis & Kirschling, 1992), some difficulties in recruitment did occur at the pediatric hospice program. It appeared that some families were not offered the opportunity to take part in the study because staff considered them too stressed to participate. Yet, parents told the researcher that only families themselves had the right to choose what they did and did not participate in. Parents held strong beliefs that staff should not make those kind of decisions without consulting the families.

While recognizing that participation in palliative care research presents potential problems, automatically disqualifying terminally ill patients and their families from research may be viewed as paternalistic (Mount, Cohen, MacDonald, Bruera, & Dudgeon, 1995). Mount et al. argue that restricting someone's rights to take part in research is devaluing and being disrespectful of that person. In addition, it suggests that they are incapable of autonomous decision making, participating in society, giving to others, or finding purpose and meaning (Mount et al.). Bruera (1994) suggests that, at least with adult patients, 80-90% of palliative patients and their families who are approached to take part in a study may consent. However, there is a perception and belief that clients should be protected from the potential harm of research, especially where children are involved. Yet, this protectionism silences the voices of vulnerable people (Aranda, 1995). As found in this current study, researchers suggest that participants often consent so that they can give something back and help others in similar circumstances (Aranda; Kristjanson, Hanson, & Balneaves, 1994). Moreover, even though the trauma associated with talking about dying experiences may be painful, it may not be harmful. In the study by Aranda, some patients and their families were upset and crying. Yet, they wanted to continue in the research because it gave them a chance to talk about a loved one when no one else was willing to listen anymore. The parents in this current study sometimes reacted in the

same manner. The potential value of participating in research should not be undervalued.

Furthermore, the participants in a study act as teachers to the researcher, and ultimately to other health professionals. They are teaching lessons that cannot be learned otherwise.

Changes within the pediatric hospice program, including changes in contact personnel and establishment of a Research Review committee after the research had been approved and initial data collection had started, and misperceptions among the management team about research with palliative care families slowed recruitment. It was never clear to the researcher how many families in the program met the study criteria as the numbers shifted over time. As best as could be ascertained, two families were asked to participate, but refused because they were too busy. In addition, two other families left the program during the study period so the staff would not approach them. There are no available data on how many other families were not approached by staff.

At the request of the staff and despite the researcher's desire for consultation, families who met the eligibility criteria for the study were identified solely by the clinical staff. Initial contact with potential participants was made by the personnel who identified the family as suitable for the study. The nurse explained to the child's parent that a nursing study was in progress, and that agreeing to talk with the researcher did not commit the family to participating in the study. The researcher was clearly differentiated from the care provider, and verbal consent was obtained to allow the nurse to forward the family member's name and telephone number to the researcher.

The researcher first contacted parents by telephone. After the study had been explained to the parents, they were invited to participate in the study. All family members were encouraged to participate. They were informed regarding the protection of their rights, and the decision to participate was entirely their own choice. When parents agreed to participate, an appointment was made for interviews and participant observations. The researcher asked the parents to

suggest the most appropriate time and place for her to come and observe them as they managed their daily lives. All parents wanted the interviews and observations to be done in the family home. Some parents were interviewed on the same day as their spouse, while others were only available on separate days. The family interview occurred after the individual members' interviews were completed.

At the first appointment, the process of informed consent was explained again. The participants signed duplicate copies of a consent form, and retained one copy. Children could not legally give consent. Provincial laws specify a legal age of consent, usually with some exceptions, and competence is determined based on maturity. Therefore, parents signed a consent for minor children. However, children who were able to communicate also needed to give assent for participation. All of the ill children and one of the siblings were unable to communicate. The researcher obtained assent from all siblings who were interviewed, although the parents legally signed the consent. Assent meant that the sibling showed understanding of the purpose of the research, knew what she could expect and what was expected of her, and was willing to participate. Consent was obtained from parents for observations of the children who could not be interviewed.

When consent had been obtained, the researcher conducted in-depth, open-ended interviews with families to obtain the participants' perspectives. Each family member who agreed to participate was interviewed individually, and then the family was interviewed as a group. This order of interviews allowed family members to say what they wanted in confidence before meeting in the larger group (B. Davies et al., 1995). The researcher was also more prepared for anything that might come up in the group interview since she could anticipate areas of potential difficulties for individuals. There is little guidance in the literature for how to approach multiple sources of qualitative data in family research. B. Davies et al. assumed that all members' perceptions of the situation were equally relevant and contributed to the whole picture of the

family, whether or not similar information was obtained in individual and group interviews. A similar assumption was made in this study.

Confidentiality can be difficult to maintain when case studies and direct quotations are used in reports, so participants were told about the small risk of public disclosure (Larossa, Bennett, & Gelles, 1981). All interviews ended with a question that asked the participants what it had been like for them to take part in the interview. Interviews lasted from a half hour to three hours, depending on the age, physical, and psychological condition of the participants. Demographic information was collected from each informant to aid in describing the sample. Interviews were tape-recorded and subsequently transcribed. The researcher transcribed the majority of the interviews.

Individual family informants were asked to tell the researcher about their experiences when a child has a neurodegenerative, life-threatening illness (NLTI). Both current and retrospective information was sought. The interviews began with an open-ended prompt such as, "Tell me about your life since (child) was diagnosed with (an NLTI). What has this experience been like for you?" The purpose of this approach was to elicit the informant's perspective with as few prompts as possible. Other broad, open-ended questions that guided the first interviews included, "Tell me about a typical day. How does your day start? What happens next? Is a typical day today the same as it was a month (or year) ago?" "What is the same?" "What has changed?" "What has helped you care for (child)?" "Can you tell me about times when something or someone has made it easier." "What has made it difficult for you to care for (child)?" "Have there been times that have been particularly difficult?" "Can you give me an example." "What is life like for you now?" One young sibling provided some data by drawing a picture. The researcher then asked her about the meaning of her picture. This discussion was recorded in field notes and on audiotape. This technique has previously been successfully used with children as young as three years and ten months (Dowden, 1995).

In family group interviews, the focus was on the family unit. Questions, therefore, were reworded to reflect the family experience. Again, both current and retrospective information was sought. At the beginning of the family interview, families were reminded about the focus of the individual interviews and were advised that some of the questions in the family interview were similar to those in the individual interview. However, the focus of the individual interview was on how the individual was managing the situation and how the individual saw the family managing. In contrast, the family interview was to determine how families managed these kinds of situations. Families were advised that some of the responses in the family interview might be the same as in the individual interviews, but other responses might be different. Questions included, "Tell me about your family's life since (child) was diagnosed with (an NLTI). What has this experience been like for your family?" Other broad, open-ended questions that guided the first interviews included, "Tell me what you do together as a family." "Is this different from before (child) was sick?" "What is the same?" "What is different?" "Why might that be?" "What has helped your family as you live with (child's) illness?" "Can you tell me about times when someone or something has made it easier." "What has made it difficult for your family to care for (child)?" "Have there been times that have been particularly difficult?" "Can you give me an example." "What is life like for your family now?" As the data analysis progressed, interviews with individuals and families became more specific. Questions were then developed depending on the theoretical requirements of the study.

Observations of the family were also used to get to know the family better. Observations were both supplementary and complementary to interviews and were made before, during, and after the interviews. These observations of interactions, such as among the family members or between the family and care providers, lasted for half an hour to three hours. The researcher observed families on between one and three occasions, depending on the families' wishes. Participation in family activities was generally inappropriate as most of the activities revolved

around providing care to the ill child. However, where possible, the researcher joined in family activities. For example, she shared lunch with one family, walked to school with another family, sat on the floor with one family as the parents played with their children, and observed and spoke with other people who were present in the family home. The influence of the researcher on families was minimized by the sheer volume of activities that families needed to perform. Families were usually so involved in caring for their child that the researcher's presence was secondary and seemingly insignificant.

Observations focused on how families managed the care of their child while also managing their daily lives; what supports were available to families; how effective those supports appeared; and how care was provided to these families. In addition, the researcher asked questions and carried on informal brief conversations with families and others following each interaction to clarify, validate, and extend observations. Other people involved in these interactions included visiting nurses, care attendants, and child care workers. Verbal rather than written consent was obtained from each person who was observed but not interviewed. Brief field notes were made in each setting. These notes were expanded and recorded on paper or audiotape as soon as possible after the observation period. Full, expanded or transcribed notes were entered into a computer program for data management. As the data analysis progressed, observations focused on checking emerging concepts from the data. An attempt was also made to negate, validate, or extend those concepts.

Interviews and observations of families can be particularly difficult because of aspects of the family that create special circumstances. Larossa et al. (1981) noted the importance of informed consent and the risk/benefit equation in qualitative research involving families. Since it is never possible to design a totally risk free study, nor is it usually possible in qualitative research to be fully aware of the complete nature of the project or potential outcomes, one should view consent as an ongoing process. Therefore, the researcher asked each participant at every

observation and interview if he or she wished to continue with the study or to withdraw with no repercussions. Consent was obtained on an ongoing basis at each stage in data collection.

Informed consent is also difficult to obtain because the pervasiveness of family life means that the range of salient issues will be extensive, and that seemingly unrelated, and often unanticipated, issues may merge during the study period. Thus, the researcher cannot forecast the exact nature of the research. Moreover, family life also has a certain inaccessibility because one cannot become a part of the family, so interviews may be the most feasible and most efficient method of collecting data from families. However, the unstructured nature of interviews can almost coerce respondents into answering, even if they would not normally talk about a subject. Conjoint interviews can be especially difficult, since they may provide less opportunity to exercise informed consent (Larossa et al., 1981). The researcher was sensitive to these issues, and interviewed each participant individually prior to conjoint interviews. Hence, the researcher became aware of issues that one or more of the family members did not wish to talk about in the group, and could guide the interview away from those areas to protect the family member.

The natural setting of the home may provide a rich context for research, yet the informality of the setting can also encourage friendliness, trust, and self-disclosure. The researcher may be treated as a guest and people may be lulled into doing and saying things they do not want recorded in a study (B. Davies et al., 1995; Larossa et al., 1981). Serendipity can also play a large role with resultant questions about invasion of privacy and consent. Additionally, the researcher is in a position of power, and a power-dependency relationship may develop, or a skilled interviewer may encourage vulnerable people to answer questions that would not normally be answered. Finally, the resemblance of qualitative family research to therapy may lead to role ambiguities. Participants may disclose more to "their therapist" than they would to a researcher. Both participants and field workers may be uncomfortable when role confusion is present. According to Larossa et al., the researcher can prevent or alleviate this

confusion by acting like a researcher, rather than like a friend. However, B. Davies et al. noted that clinical researchers may have great difficulty in maintaining a researcher role. Awareness of these potential difficulties, critical reflexivity, and constant examination of actions and interviewing techniques helped the researcher to reduce the risk of misuse of her position.

Data Analysis

Analyzing data of this nature is always intellectually and emotionally challenging (B. Davies et al., 1995). The interviews involved discussions about death and dying in children and portrayed family disruptions that caused the researcher to experience sadness and pain. The researcher needed opportunities to talk about and to deal with the emotional impact of the data. Her dissertation supervisor, and to a lesser extent her dissertation committee, provided a source of support when needed. In addition, the researcher discussed her experiences with colleagues who have conducted research in sensitive areas. B. Davies et al. noted that research of this nature should not be a solitary endeavour because of the potential for emotional reactions to the data. Therefore, the researcher analyzed the data in collaboration with her dissertation supervisor. This collaboration was an opportunity to gain greater insight into the data because of dual perspectives in analysis. In all discussions, anonymity of the participants was respected.

Management and analysis of transcribed field notes and interviews were facilitated by the use of a word processor computer program. Each transcript was entered sequentially, and all interviews and observations pertaining to a family were kept in unique directories. Transcripts were coded directly on the computer, and codes were embedded in the transcripts using a bold style to facilitate easy compilation of a code list. In addition, data could be searched for particular codes in different transcripts simply by using the "Find" feature in the word processor program. Sections of data from different transcripts could then be copied to a new file for ease of retrieval. Code lists were dated and were compiled for every transcript, and for each time a transcript was coded. Codes were linked to the appropriate transcripts. Each recoded transcript

was dated and numbered to facilitate retrieval. Over time, code lists were merged and used for further analysis. All prior code lists were retained in order to keep track of the development of codes and categories. Memos were written whenever the researcher was theorizing or worrying about anything. Participants' code numbers and the date were included on each memo. The researcher wrote out her thoughts and included the code names that she was thinking about when she felt the need to write a memo. All memos were carefully linked to the appropriate transcript. As data analysis progressed, the researcher drew diagrams to capture the relationships between categories and subcategories. Hard copies of all transcripts, code lists, memos, and diagrams were kept in binders for easy access during data analysis.

Data analysis was concurrent with data collection, since analysis guided the questions for future data collection. Theoretical sampling decisions were made on an ongoing basis, depending on the needs of the study. For example, as data were analyzed, the researcher sought to interview families who did not live in Vancouver, were non-Caucasian, had more than one ill child, had siblings, or lived in varying socioeconomic circumstance. The coding procedures laid out by Strauss and Corbin (1990) were used to analyze the data. These procedures build, rather than test theory. They give rigour to the process, help break through the researcher's biases and assumptions, and provide the grounding, build the density, and develop the sensitivity and integration needed to generate a rich, tightly woven, explanatory theory that closely approximates the reality it represents (Strauss & Corbin). The researcher moved between one type of coding to another in a given coding session, especially from open to axial coding, and continually asked many and diverse questions about the phenomenon under study.

Constant comparative analysis of each transcript and field notes was used to check for commonalties and differences both within and across transcripts. Data from individuals were compared and contrasted with both similar and dissimilar transcripts from individuals, such as a mother being compared with another mother and contrasted with a father. Group data were

compared with other group data, and were also contrasted with individual data. In addition, transcripts involving one family (observations, individual family member interviews, and the group interview) were compared and then contrasted with data from other families. Thus, commonalities and differences within and between individual and family data were discovered.

"Open coding" was the first level in data analysis (Strauss & Corbin, 1990). Open coding is the process of breaking down, examining, comparing, conceptualizing, and categorizing data. Initially, open coding was non-judgmental and only named the ideas that were in the data set. Each transcript was broken down into units and open coded two or three times, at least in the early stages of analysis. Transcripts were examined line by line to identify codes or words that captured the meaning of the events (Strauss & Corbin). Once a number of transcripts had been open coded a few times, the researcher conceptualized the data by giving similar phenomena the same conceptual name. There were many conceptual labels, so these concepts were then grouped into preliminary categories to reduce the number of units with which the researcher needed to work. The conceptual name given to a category by the researcher reflected the data that it represented. Questions were continually asked of the data to develop categories in terms of their properties (characteristics or attributes) and dimensions (locations of a property along a continuum). This process formed the basis for making relationships between categories and subcategories, and later between major categories.

The next level of analysis involved "axial coding" where the researcher made connections between a category and its subcategories (Strauss & Corbin, 1990). Categories were linked and developed by asking questions and making comparisons. The researcher asked questions about the type of relationship, in terms of the conceptual labels, and looked at the data for evidence, incidents, and events that supported or refuted the questions. Evidence of differences and variation added density and variation to the theory. The researcher continued to look for other properties of categories and the dimensional location of each incident that was coded. Patterns in

the data were noted to provide the basis for selective coding, and concepts and relationships were constantly verified with the data.

A coding paradigm (Strauss & Corbin, 1990) was used to specify a category (phenomenon) in terms of the conditions that gave rise to it: causal conditions, context, intervening conditions, action/interactional strategies, and resultant consequences. A phenomenon is the central idea or event towards which a set of actions/interactions is directed to manage or handle it, or to which the set is related. In order to discover a phenomenon, the researcher asked questions such as, "What is it that these data are referring to? What is the action/interaction all about?" Causal conditions are the events or incidents that lead to the occurrence or development of a phenomenon. Words such as when; while; since; because; due to; and on account of were often cues to causal conditions, although the researcher sometimes needed to check back from the phenomenon to find the causal (antecedent) condition. The context provides a specific set of properties that pertain to a phenomenon and shows how the phenomenon is handled under a specific set of perceived conditions, whereas intervening conditions are the broader structural context pertaining to a phenomenon that act to facilitate or constrain the action/interactional strategies taken within a specific context. These conditions need to be managed. Action/interactional strategies are purposeful and goal-oriented, and are done for some reason in response to or to manage a phenomenon. Words to look for included action-oriented verbs or participles. Finally, consequences are the outcomes of the action and interaction. Consequences were events, happenings, or responsive actions/interactions that were actual or potential and that occurred in the present or in the future. The consequences of one set of actions sometimes became part of the conditions in another sequence.

"Selective coding" is the process of selecting the core category (central phenomenon around which all the other categories are integrated) by systematically relating it to other categories, validating those relationships, and filling in the categories that need further

refinement and development (Strauss & Corbin, 1990). This is not a linear process, since the researcher must move back and forth between "steps." Selective coding is similar to axial coding, but occurs at a more abstract level. At this level, the researcher tried to identify the story by writing a memo that captured the essence of the story in a few sentences. Then the researcher moved beyond description to conceptualization and told the story analytically.

The central category was named with a high level of abstraction, and the chosen conceptual label fitted the story it represented. Questions that the researcher used to identify the central category included, "What phenomena are reflected repeatedly in the data? How can the findings be summarized? What essential message about this area does the researcher want to pass on to others? What can be considered important about this area and why?" The core category was developed in terms of its properties and then dimensions. All other categories were then related to the central category in subsidiary categories by means of the paradigm: conditions, context, intervening conditions, strategies, and consequences.

When certain categories were consistently emerging and no new information was being discovered, then theoretical saturation was reached and data collection ceased. However, refining the theory continued into the writing phase and necessitated the researcher returning to the participants to collect further data to fill any gaps that existed in the theory (Strauss & Corbin, 1990). The developing theory was grounded by validating it against the data. The researcher looked for a general fit, not necessarily a perfect fit in every case, and modified the theory until a general match was made. When the theoretical framework held up to scrutiny and the conditions and processes were built in and accounted for, then the researcher went back to the categories and filled in any missing detail to give conceptual density and increased conceptual specificity.

During development of the theory, the researcher also attempted to identify the process that was involved in the families' experiences by linking action/interactional sequences. Retrospective data obtained from families were used in this analysis. The researcher noted

changes in conditions that influenced action/interaction over time, the action/interactional responses to that change, and the consequences that resulted from that action/interactional response. She then described how those consequences became part of the conditions influencing the next action/interactional sequence. Process is often a difficult idea to grasp, because identifying process involves an in-depth examination of and incorporation of changed action/interaction into the analysis, as this varies over time and in response to changes in conditions. Because process explains growth and development, or the lack thereof, then the researcher needed to account for or explain any variation in the rate of movement, or why a passage downward might reverse itself or be interpreted, and with what consequences.

Analysis was aided by the use of memos. In grounded theory, memos have a specialized meaning - they are the written records of analysis related to the formulation of theory, and they represent the researcher's abstract thinking about the data (Strauss & Corbin, 1990). Memos help the researcher to move away from the data to abstract thinking, and then to return to the data to ground the abstractions in actual data. Memos also show the theory developing step-by-step, and are crucial in developing a dense, well integrated theory. Memo writing began at the inception of the research project and continued until the final writing. Different types of memos were used to facilitate analysis. Memos related to codes contained the actual products of the three types of coding, such as a category with some of its properties and underlying dimensions. Theoretical memos were theoretically sensitizing and contained the products of inductive or deductive thinking about categories, their properties, dimensions, relationships, and variations (though they were provisional until validated with data). Operational memos contained decisions and directions to the researcher regarding sampling, questions, possible comparisons, and leads to follow.

Scientific Rigour

Qualitative research must be rigorous, but does not use the same criteria of scientific rigour as quantitative research. Criteria include truth value or credibility, applicability or fittingness, consistency or auditability, and neutrality or confirmability (Hall & Stevens, 1991; Sandelowski, 1986; Schutz, 1994). Rigour was maintained by following the procedures of grounded theory (Strauss & Corbin, 1990). Credibility, fittingness, and trustworthiness of the data were ensured by constantly checking to ensure that the coding categories portrayed the data as a whole, and by checking with families to ensure that the interpretation made sense to them. In addition, presentation of findings to colleagues in the field provided further support for the theory. Theoretical sampling ensured the relevance and comprehensiveness of the data, and adequacy was obtained by assuring that all categories were saturated. Auditability was facilitated by consistency in data collection, format, and analysis, and in consistent routines for recording and reflection. The development of the analysis is demonstrated in this final written research report that reflects the complete research process.

Ethical Considerations

Ethical clearance was sought and obtained from the University of British Columbia, Behavioural Sciences Screening Committee, and from the board and or ethics committee of each of the facilitating institutions. Each informant was given an explanation of the study. They were told that their participation was voluntary and that they could withdraw from the study, refuse to answer any questions, or terminate interviews or observations at any time. Participants had an opportunity to ask questions before consenting to join the study. Written informed consent was obtained from each participant. In the case of minor children, parents signed the consent form, but children who were cognitively able also gave their assent to participation. In addition, ongoing consent was obtained verbally at each interview or observation. Informants were assured that all information would be confidential, and only the researcher would have access to

the identifying information. Data were kept in a locked cabinet that could only be accessed by the researcher. Selected portions of the data were shared with the researcher's dissertation committee for purposes of illustration and analysis, however, anonymity was preserved.

All participants were asked if they wanted to receive any of the results, and in what format. Some chose to review their personal interview transcripts, while others requested a summary article. All parents asked for a copy of the final research report. Transcribed interviews were mailed to participants who requested a copy of their own interview. No individual's interview was given to another member of the family. Each family member's interview was mailed in a separate envelope. The envelope was addressed to one particular individual and contained only that individual's transcript. Parents confirmed that confidentiality would then be maintained. All identifying information was removed from transcripts, field notes, and demographic information. Code numbers were assigned to each participant. The master list of code numbers and names was kept separately from the data in another locked drawer that was accessible only to the researcher. At the conclusion of the study, all identifying information and audiotapes will be destroyed. Transcribed data will be retained for educational and other research purposes, subject to further ethical approval according to standard procedures.

Summary

This research was designed as a grounded theory study that focused on the experiences of families with a child who has a neurodegenerative, life-threatening illness. A symbolic interactionist perspective guided the method. The study was conducted in a natural setting; in the homes of families who agreed to participate. Interviews, supplemented by observations, were used to enhance understanding of the experiences of families with a child who has a neurodegenerative, life-threatening illness. Every member of the family was encouraged to participate. Interviews were conducted with individuals, and then with the family as a whole. This study generated a detailed, contextually-grounded description and theoretical explanation of

the experiences of these families. The knowledge generated by this study and its contribution to developing theory will be used to promote effective palliative care of families with a child who has a neurodegenerative, life-threatening illness.

The grounded theory that evolved through interpretation of the data in this study will be presented in the following chapter. In chapter six, the process that families went through will be discussed and placed within the context of the current research and literature. The study will be summarized in the final chapter, and implications for practice, education, and research that arose from this study will be identified.

Chapter V: Findings

Navigating Uncharted Territory: Experiences of Families When a Child has a Neurodegenerative, Life-Threatening Illness

The purpose of this chapter is to present the grounded theory that evolved through interpretation of the data in this study. This grounded theory of the experiences of families when a child has a neurodegenerative, life threatening illness (NLTI) is the product of multiple phases of analysis. The basic social process of *navigating uncharted territory* (see Figure 1) characterized the process that these families went through. *Navigating uncharted territory* was comprised of four main dimensions: *entering unfamiliar territory*; *shifting priorities*; *creating meaning*; and *holding the fort*. Each dimension will be explored in-depth later in this chapter.

Experiences do not occur in isolation. Instead, they occur within a sociocultural context that can influence and constrain a person's ability to act in certain ways. Explicating the context of an experience is very important therefore, because it provides a framework within which the experience can be understood. Consequently, the context of the illness experience will be described prior to presenting the grounded theory. In addition, certain intervening conditions can inhibit or facilitate the strategies people use within a specific context. Four intervening conditions that moderated the families' experiences in this study, *relationships with health care providers*, *availability of information*, *gender differences*, and *communication between parents*, are described following presentation of the theory. Discussion of the findings and their relationships to current literature will take place in a subsequent chapter.

Context of the Illness Experience

The onset and diagnosis of a child's neurodegenerative, life threatening illness (NLTI) affected not only the child, but also enveloped the whole family in the illness experience. Moreover, the illness experience evolved in and was created by the interaction between a

Acute, curative health care system - Sociocultural environment

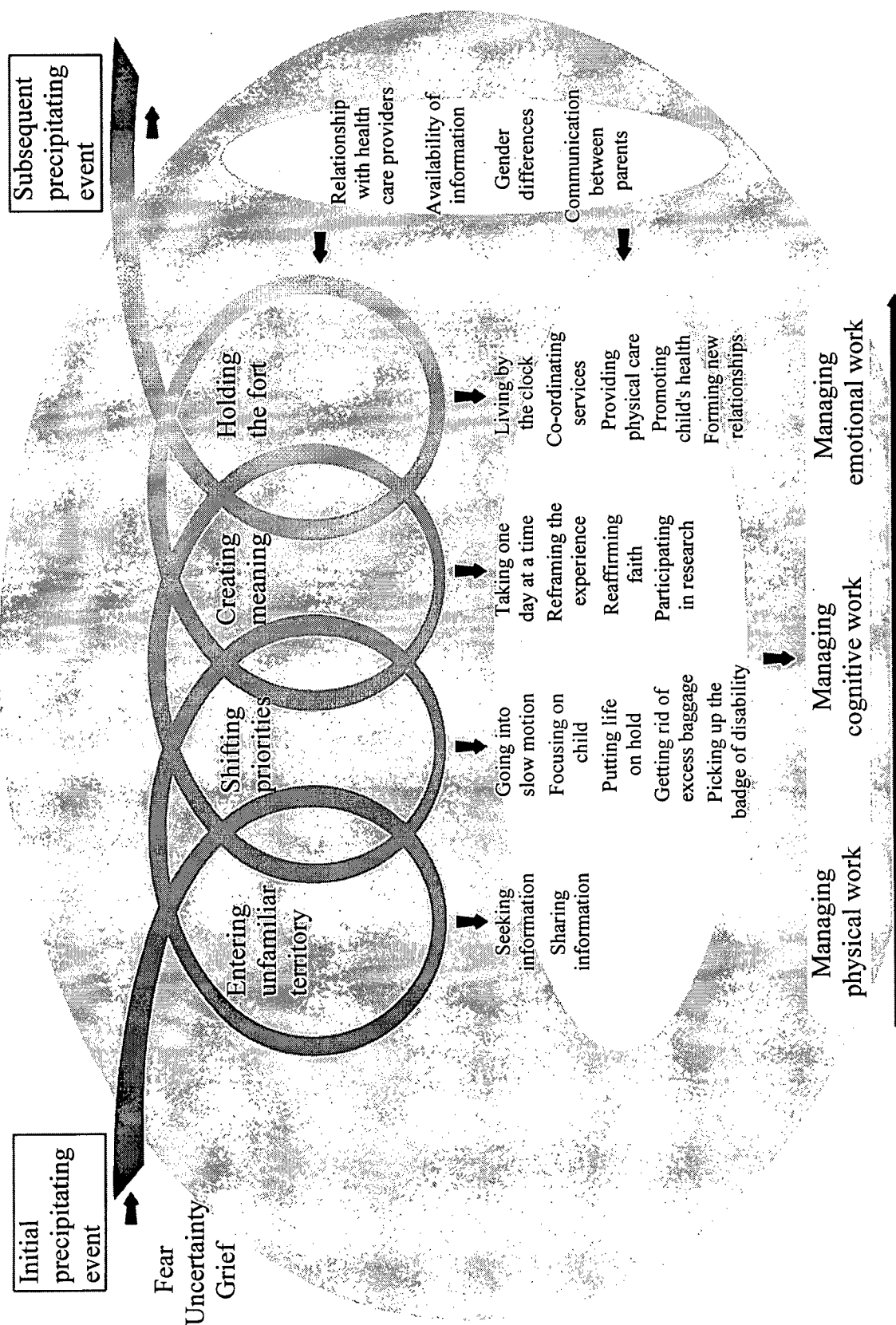


Figure 1. Navigating uncharted territory

specific disease process and the family's social world, particularly the health care system. Yet, few parents had thought about the system until they became involved in it. They had assumed that the system would be there to support them if and when they needed it. Reality was usually very different from their expectations. Most parents were frustrated by what they perceived to be a complex social institution. Confrontations with the health care system were endemic. The context of the illness experience encompassed two main areas, the *acute, curative health care system* and the *sociocultural environment*.

Acute, Curative Health Care System

Care was provided to the children within an acute, curative model that was, from the perspective of families, inadequate in meeting the needs of families where a child had an NLTl. Families were adamant that "the whole family is affected when a child is ill," and that family-centred care is critical. Yet, healthcare professionals frequently neglected or failed to recognize the impact on the family. Their interventions typically focused only on the child.

The system structures were designed to provide emergency and highly technological services. They emphasized expensive technology to discover new curative procedures. However, these children could not be cured. Parents commented that their children and families "fell through the cracks in the system" because little money and attention were available for chronic, incurable conditions. Parents perceived that, unlike illnesses such as cancer, few people knew about neurodegenerative diseases. In addition, allocation of resources was viewed as "almost like politics or a vote." One parent explained, "When you get the most votes, you win. If you have cancer, you win because more people have cancer than NLTIs." In addition, there were few organizations or camps that would assist the children because their diseases "did not fit the criteria" set for funding or admission to programs.

Even though there was no available cure and limited treatment, families were often still caught up in a whirlwind of health care involvement. For example, immediately following the

diagnosis, physicians were sometimes anxious to try innovative research protocols, such as experimental bone marrow transplants, in a bid to slow down the progression of the disease. However, parents reported that they were not always given full and accurate information with which to make informed decisions about such treatments. Some parents resented the expectations of their health care providers that the family should participate in medical research and education. They stated emphatically that their children were not "lab rats."

Other health care involvement occurred because children with NLTIs often required diagnostic testing, home support services, ongoing therapies, or frequent hospitalizations for acute illness episodes. Yet, families perceived that their children were vulnerable to error and incompetence on the part of health care providers. Parents described their horror, fear, and frustration resulting from perceived incompetence that was sometimes compounded by the health care provider's unwillingness to listen to the parents' warnings. In a few families, parents felt that their children's quality of life had been compromised by incompetence. Moreover, they believed that the acts were not regarded as incompetence within the medical establishment. Sadly, parents were often not satisfied that other families would not be treated in the same way in future. One mother described a rather extreme case involving her son who had returned to the ward following treatment in the Intensive Care Unit (ICU):

They had Ian on oxygen. [Staff from] ICU would come up and reduce the level of the oxygen every day. She [child's primary care nurse] came up after four days of not seeing Ian. She told us how wonderful Ian looked. She then removed the oxygen. Sam [husband] and I were standing there going, "Don't remove the oxygen. The oxygen is being lowered by ICU. Don't remove it." She removed it. An hour later, Ian had a seizure and a stroke.... Of course, when you ask doctors if he had his seizure because of the oxygen, "Well we don't know that. We don't know that." So, I was really getting frustrated with that answer. I phoned our pediatrician here in [city] and said I want a straight answer. He said, "Definitely. That's exactly what happened to him. But Linda, you're asking these people to accept liability. It is not going to happen." I said that is NOT RIGHT. That is not right. They made a grave error with our son. And she [the nurse] was fired. She told us, they didn't tell us that. I heard rumours that she was going to Ontario or something. She met Sam and I in the hallway and she was as white as a ghost. You could see that she was very upset. She told us that she had been fired. This [child's seizure and stroke] shouldn't have happened.

Parents were frustrated when, though in theory they had access to care, they did not find the support or assistance they needed. In addition, they often had no individual health care provider as a primary resource so parents felt adrift and alone. Isolation was more common when families had few links to the hospital or were not receiving services at home, often because the child was not yet sick enough to qualify for home support. Feelings of isolation after leaving the hospital were described by one mother this way:

And the other thing that I really don't like and really, really, really frustrates me is that we went into the hospital and everybody had all these wonderful goals and wonderful ideas. But once we got him out of the hospital, it was BOOM. That's it, you're cut off. No conversing, nothing. We go back once a year. That's it. Nothing. Nothing. You're talking about feeling abandoned. That's unbelievable.

Moreover, it was seldom that professionals offered information or resources. The onus was usually on families to "go out and find it." Parents spent a lot of time and energy "running all over the place." They felt that having the sole or major responsibility for figuring out information and resources was "too heavy a load." In many cases, they accidentally discovered information and resources to which they were entitled. This father's experience was common:

I didn't know about the government programs. Even when I went on welfare and told them that Cory was sick, nobody mentioned anything to me. It was my mum. She joined the [child's illness] society. One of the ladies she talked to said, "You shouldn't be paying all this money for prescriptions and things." That's how I found out about the programs.

Parents also found it difficult to challenge the rules and regulations of a highly bureaucratized system. The regulations often seemed to be illogical. For example, the provincial system would pay for a child to be placed in an institution at a cost of tens of thousands of dollars per year. But it would not pay for in-home care at a fraction of the cost. Some parents suggested that the system was set up in a way that created and maintained jobs for unionized health care providers in hospitals.

Most of the child's care was provided at home. Therefore, parents were heavily involved in lobbying for, obtaining, and co-ordinating support services in the home. However, the rules

and regulations governing home support services were particularly strict and frustrating in the eyes of parents. One family, who wanted respite services, was told by a nurse from their local home care program, "But this is our policy. Unless he needs naso-gastric tube feeding, you get nothing." The number of hours and frequency of home support were decided by the program staff. There was little flexibility in changing the set hours provided. For example, if the scheduled night nurse was sick, families neither received a replacement nor an option to use the hours at an alternate time. Parents felt that they were expected to function without sleep. One mother of two affected children stressed the value of being able to hire private duty nurses:

The government program tells you when you can have a nurse. And the nurse can only work with Sarah because Nancy is not classified as sick enough for a nurse. With a private nurse, you can set your own hours. If they cannot work the hours you need, then you don't hire them. They will also work with both Sarah and Nancy because that is what I hire them for. It's really important to have the flexibility that you can't get with the government program.

However, not all families could afford the luxury of hiring private nurses. This mother could only afford to do so because of the extended medical benefits available through her employer. If her company had forced her to resign instead of putting her on long-term disability, then she would not have been eligible for the private nursing care.

Parents identified inequities in the system that were often related to where a family lived. There was a lag between needing services and receiving them that increased when a family did not live in a resource rich area. In addition, families often received very different services depending on where they lived. Most of the resources were located in the Vancouver vicinity. But some families could not afford to live in Vancouver:

There haven't really been any resources. Then the couple [of resources] that have been created along the way, they're too far for us to attend. But if you live in North Vancouver, there's plenty there.... The Neurological Society in North Vancouver and their building and their facilities. But Ian can't take advantage of that because we live [out of town]. I couldn't afford to move into Vancouver if my life depended on it.

The vagaries of the home support program also prevented some families from moving, because they were satisfied with existing services. Some families turned down opportunities to relocate with a new job. A father explained:

If we go somewhere else [still in the same province], we have to start all over. With the government arrangements the way that they are, home support is now a year's wait. There's a year's wait for this, there's a six month wait for that. So, you could be waiting a long time for the services that we already have. And we would lose some [services]....Different district, different person [social worker], different responsibilities.

Many parents faced territorial battles over which treatments and therapies would be supported by the health insurance program. For example, one child required an expensive treatment that was provided for free when he was in hospital. His parents were told that "as long as he's in hospital they have to give this to you." But, the parents were required to pay for it when he was at home. They could not afford the high cost. So, staff would not allow them to take their child home. These parents, like some other parents, aired their difficulties in the media and also contacted their local member of parliament. Eventually, the treatment was funded in the home and the child could go home. Strategies such as using the media or politicians required determination and assertiveness. Not all families had the ability or energy to continue fighting in these ways for what they wanted.

Sometimes, a family's case fell between a number of government agencies. No one took primary responsibility for the family. In such cases, the onus was again on the parents to take the initiative and to find ways to obtain assistance. One father noted that his family only received the respite assistance they needed because he and his wife organized a meeting with pertinent personnel:

It wasn't until we met with an inter-ministerial committee which had people from the Ministry of Health, Ministry of Social Services, some other Ministries as well. I actually prepared a written brief about what we wanted. We had documentation...letters from different physicians, pediatricians. We also had social workers there to speak on our behalf....It took a few hours to put everything together. The social workers were very helpful in making sure we had the right channels. It was a matter of sending a few letters here and there and then actually going to this meeting.

Families also incurred many financial expenses that were "hidden costs." All families received some financial support from various government programs. Numerous costs, however, were not covered. These costs could be quite substantial, especially for families whose incomes were diminished because of the child's illness. For example, many children were unable to regulate their internal thermostat adequately or required warmth because they were susceptible to illness when they became cold. Consequently, the room temperature was often kept high, resulting in excessively high heating bills. One family needed an air-conditioner in the child's room during the summer time because of her sensitivity to heat. Extra laundry, because the child drooled constantly or was incontinent or vomited and soiled the linen, also added to electricity costs. Most children were incontinent and required diapers which were expensive. Government programs considered diapers a normal expense for families with infants and young children. They only covered diapers when the child still required them beyond the age of three years.

It was virtually impossible for these children to wear regular clothes. Their diseases caused abnormal physical development. Specially made clothing could cost hundreds and even thousands of dollars over the years. Children often needed equipment, such as wheelchair ramps, in order to get out of the house, or wheelchair lifts costing \$5,000 to assist parents to lift the child and wheelchair into the van. A van cost \$30,000 by the time it was adapted for the wheelchair. These expensive items were not covered by government programs. Families had to look for funding elsewhere. If families could not find extra funding, they usually had to do without the item because they could not afford to pay for it.

Most children required tube feeding. The government programs usually paid for commercially prepared enteral feeds. However, many families cooked special diets, such as high protein foods that were quite expensive. This added expense was not covered. Some children could swallow commercial baby food, but the government did not cover this cost either. While most, although not all, prescription medications were paid for, these children usually required

other drugs such as vitamins, non-prescription analgesics, or anti-pyretics on a very regular basis. The cost of these additional and necessary drugs could become very high. Families sometimes had to do without other things in order to buy the child's fever medication.

When children were hospitalized, sometimes for 10 days every month, costs to families ranged from \$40-50 a day for food, parking, gas, coffee, magazines, and so on. Although parents tried to limit such expenses, the minimum was still \$40 a day month after month after month. In addition, when they were at home, parents often had no time to cook or they would be so exhausted that they would order food in. Again, food was an added expense that became part of the family budget.

Even the cost of a rare evening out was more than would be typical for families with only healthy children. In such families, grandma could baby-sit for free or maybe the teenager next door would charge \$5 an hour. But when children had complex needs, families needed a specially trained child caregiver or even a registered nurse. Hourly child care fees then became \$12-20 per hour or even higher.

Families with lower incomes barely survived financially. They talked about living from day to day, just able to pay their bills, not really living but only "existing." As one father said:

We are able to just cover and make our ends meet. But any major financial crisis that comes about, we've had it. We cannot afford to buy a van or anything like that, but it is fine. Also, the other thing is when you go through something like this, you learn very quickly to deal with one day at a time.

In families with higher incomes, parents often felt guilty when they accepted government or charity assistance. However, they were very clear that the financial assistance allowed them to continue caring for the child "without burning out." They needed a break and they used the financial assistance to get that break. These parents talked about how hard it would be if they could not enjoy some pleasures in life.

Sociocultural Environment

The behaviours and attitudes of extended family, friends, strangers, and health care providers also influenced families' lives. Moreover, the sociocultural issues were often reflected in the encounters that families had with the individuals who provided services within the health care system.

Health care providers.

Parents were often shocked to discover that health care professionals exhibited some of the same prejudices that were present in the general public. For example, less privileged people, such as those on welfare, faced considerable prejudicial attitudes from some health care providers. This prejudice was a source of great fear for parents and was very disturbing to them. One mother lobbied to remove explicit indicators of this stigma:

They [hospital staff] found out we were on Social Services. Then we were down on that totem pole. I tell you, we were nothing from then on. And that offended me. That's nobody's business but my business and my husband's business. That's the part that angered me the most. I got to the point where I even said that to a nurse, "With me being on Social Services, you're guaranteed payment lady. So back off." And that's why the medical system now has changed. On your medical card it no longer says "H2" where that would indicate that I'm on Social Services. It no longer says on his [child's] health card "S2" which means he's on the government program. That's no longer there.

One unemployed father reported an unfair assumption by health care professionals that he was taking illicit drugs. This experience was very upsetting for the family and coloured their future relationships with other health care providers. He described what happened when he went to the Emergency Room of a major hospital:

I said, "Look, I feel like I'm going to explode. I haven't slept in four or five days literally. My anxiety level with my son and stuff is super high and I'm scared of what I might do." So, they put me behind this thing. "Take off your clothes and put on this gown." Then I had this doctor come up to me. He was treating me like I was some kind of drug addict from the street that was in Emergency wanting a Valium to calm down because I couldn't get any heroin or cocaine on the street. This was exactly how he treated me. Here I am, my kid's [seriously ill] two blocks down the road [in hospital]. So, literally I just put my clothes back on, told him where to go and left. I mean, "How sick!" That's ridiculous. There's no way a father should have had to go through that. And really, that's how I was treated. Like I was just looking for a fix. I know I was wired for sound, but that was just

me. I needed to calm down. A couple of the nurses gave me two children-size Ativan at Children's [hospital], because now I was just totally off my rocker after running into this yahoo [physician]. It was just ridiculous.

However, when parents were professionals, such as teachers or accountants, they were treated with more respect. Parents perceived that health care providers frequently expected parents who were professionals to be more involved in decision making. Health care providers also appeared to give more weight to input from such parents. In turn, parents who were professionals were often more able to draw on interpersonal skills, abilities, and resources from their professional lives that served to reinforce the health care providers' perceptions.

Parents also reported frequent power struggles with health care providers. They were sometimes left with an unpleasant memory that affected future health care experiences. Parents surmised that many professionals had a need to always be right and to be in control. In some instances, professionals used rudeness and intimidation tactics to reinforce the power imbalance. Some health care providers displayed an attitude that implied that independent decision making on the part of the parents was wrong. For example, the parents of a boy who was in adrenal failure wanted the physicians to give him Hydrocortisone, because this drug had been successful previously. However, some endocrinologists insisted on giving the standard treatment, Prednisone, which the parents knew from experience did not work for the child. Finally, an older physician pulled them aside and said, "I'm going to teach you how to do an intramuscular injection of Hydrocortisone. You don't tell anybody." The parents gave the injection to their child whose condition then improved dramatically. However, parents then had to deal with other consequences:

Then Endocrine [specialists] refused to see him because we introduced the intramuscular injections without their approval. They were like little children. They were mad because we had made a decision for our son. I said, "I don't care. I will give this injection every day. I don't need a nurse to do it. You don't want to do it, we will do it." And we did it. She's [endocrinologist] still angry to this day.

Extended family and friends.

At the beginning of the child's illness, extended family and friends were usually very supportive and helpful. But, over time, they often withdrew. Parents explained that people were afraid of the child's seizures and erratic behaviour, or they did not know how to communicate with a child who could no longer talk. In other cases, people stayed away because they did not know what to say to the family. Very few friends and extended family members actually remained supportive during the long course of the child's illness:

We are finding that right now people are not coming as they used to. When our son was first diagnosed, we had a lot of people coming. There was a lot of activity, people always coming and finding out about him, but now it is not as much. I think maybe people are quite used to that and they say, "Well he is still there and still going strong." Which is fine, but it is also important for them to realize that it is getting harder for us. But anyway, it is something that we have to deal with.

Families found it difficult to accept losing relationships and friendships, especially when they were losing people "that you always thought you would have." When one father asked his psychologist to explain why the journey was "getting lonely," he was told:

And the explanation I got was that people get so used to you dealing with your child. As well, for them, in the beginning, everybody pours their heart out. They will come and help through their words, through their visits, through their actions, everything. But then after a while, there is only so much they can do and only so much they can give, and they realize it as well. They also get caught up in their own cycles. So basically, I find, eventually, you are on your own.

One mother tried very hard to understand why friends and family would stay away. She made a special effort to overcome their reluctance to visit. This mother wrote a book to tell others what was wrong with her daughter and to help them understand the family's situation. She found this approach valuable:

I wrote that [book] specifically because our friends, the kids were okay, but the parents would come and they would sit and they'd be so uncomfortable. Talking got to be very difficult. We'd be talking about Amanda and they would look very uncomfortable. Then you'd think, "Okay I won't talk about it." Then they just stopped coming. The more uncomfortable they got, the more uncomfortable the kids got....I sat down and wrote the book. Then I gave them all personal little copies and it was amazing. It made a very big difference so that they started asking questions then. Or maybe they didn't need to ask the

questions anymore because the questions were answered in the book. It was like we could just now go on. It did facilitate [communication] later on, within weeks. Then they would start to say, "I didn't understand this" or "Why does that happen?" They started treating her like she was before, before the diagnosis.

However, not all parents had the strength or resources to make such an effort. Many families were disappointed and hurt at the lack of support from friends and extended family members. Another mother suggested that other people "did not want to understand because it scares them....It really, really frightens them to think that maybe one day they'll have to live the kind of life I live."

While families did not expect extended family and friends to fully understand their situation, they wanted them to know "what it's really all like" because most other people didn't really know. However, families were also wary of being honest. They did not want to impose their hardships on others or make them feel sorry for the family. In the words of one mother, "You need to be careful what you tell your friends. You limit what you tell others and only tell them what they want to hear." But, when other people indicated that they were willing to hear the truth, families were willing to be open:

Sometimes when a pointed question [is asked], it really helps because if you give a good pointed answer, it solves a lot more than if you just sort of go, "Well she is feeling pretty good today." Because when someone asks you a pointed question, then you can basically say, "No, she is doing lousy, or she is doing this, this and this." So those sorts of things really help that way.

Not only did some people not want to know the truth, sometimes they made erroneous assumptions about the family's situation. Families found that especially hard to tolerate. One father expressed his annoyance:

[The] ignorance of some people as well. Those that don't ask questions and then assume things....Even when you don't tell them what is happening, they hear by the grapevine or whatever. Those are people that don't help because they don't care to find out what is really happening. They just assume that you are unable to do what you want to do and then [they] avoid you at all costs almost.

Families felt supported when extended family or friends did something concrete like making meals for the family freezer, or helping with paperwork, painting, or sibling care. Such activities were only supportive, however, if the family needed and asked for that kind of help. Offers of help that the family did not perceive as useful were more of a nuisance than a help. Families also appreciated words of encouragement that they were doing a good job and they gained more energy to carry on.

Impact of visible differences.

Children with an NLTI were usually visibly different from other children. In many cases, the child's mobility was restricted and he/she was confined to a wheelchair. Stunted growth, twisted limbs, or other altered physical attributes were also common. Some children exhibited unusual behaviours, such as non-stop screaming for no reason or an inability to speak. Most of the children experienced dietary alterations that necessitated feeding through a tube into their stomach, or at best, they could only eat a soft pureed meal. The impact of these visible differences was profound. The children attracted immediate attention and invoked a variety of responses. Some people were sympathetic, but many others stared or were afraid of the child. One parent explained that people "did not know how to come forward. They were scared to take the initiative, afraid to ask what was wrong. They didn't know what to do." Parents noted that there was "a little bit of stigma attached to going into a function with a child in a wheelchair." Despite their best efforts at educating others, parents believed that "the look" would never disappear completely. Parents were often embarrassed and uncomfortable. In most cases, they eventually gave up attending church functions, taking the child to the movies, on camping trips, or to the sibling's baseball team barbecue.

Conversely, visible differences sometimes created an opportunity to let the world know about the child's condition without having to give continual explanations. In addition, people sometimes reacted positively when the illness was visible. For example, when one child was in a

wheelchair instead of his regular stroller, people would hold doors open for his parents and smile at him and his family.

In summary, the experiences of families as they navigated uncharted territory occurred within a context that was complex and often constraining. The families' social world, particularly in relation to the health care system and those who deliver services, provided the background in which the illness experience unfolded.

Navigating Uncharted Territory

Families living with a child who had an NLTII were looking for ways to deal with the unfamiliarity, uncertainty, and unpredictability of their lives. Most were unfamiliar at first with the world of disability, the health care system, and the range of options that were available to them. In talking about their experiences, parents spoke about being on a journey, a journey that was unlike the "normal" life journey. They tried to describe an experience that was unique, filled with constant poignancy, and "indescribable." The experience was incomparable with other life experiences, no matter how difficult those other experiences may have been. One mother said:

I've actually wrestled with trying to put onto paper what it has been like. There are just so many things, there are so many aspects that it is just impossible. What has it been like? If you wanted me to say it in one sentence, I'd say it has been the worst experience of my whole life and I hope I never have to go through this sort of thing again....I've been through a lot of other things. My mom was diagnosed being terminally ill. I've gone through...I had some sexual abuse when I was a young girl and I didn't remember it until I was 27 or something. So [I've gone] through all that trauma and everything and nothing can compare to this. Nothing.

The journey was filled with fear, uncertainty, and grief. It was a journey that some families endured and suffered through with a "heavy heart," while others looked for the positive aspects and found some good in the situation. It was also a journey that was forced on families, one they would not have chosen, and for which they could not change the final outcome. They did not know when they would reach the end of this particular journey or how they would get there. Just when families became accustomed to a fairly comfortable position on a plateau of

relative stability, the child's condition changed again and they were thrown into unfamiliar territory once more. Families were travelling without the benefit of a "map" that might be based on the experiences of many other families. Because relatively few families had gone before them, or at least had documented their experiences, these families were travelling in uncharted territory.

Parents reported a range of onset patterns for the child's illness, but it was important for them to identify a beginning point in the illness experience. For many, the beginning of the journey came when a parent, usually the mother, noticed that the child was exhibiting some unusual behaviour or symptom, such as a limb tremor or a loss of motor skills. In most families, the behaviours were acquired weeks to a few years after birth. Only in one family was the child noticeably affected at birth. A diagnosis was not made in this case though until the second affected child was born and exhibited similar symptoms. The process of diagnosis was often long and drawn out and was accompanied by severe emotional distress in most families. A major component of this distress was uncertainty and fear about the potential diagnosis. Yet, once a diagnosis had been made, most parents were relieved. As one mother recalled:

At least once you know, then you can go, "Okay, I know where I am and now I can kind of go on." You start to cope. But before that, you are just so scared all the time that I think fear is worse than knowing.

Although the children were diagnosed with illnesses that would eventually kill them, families remained uncertain about how the child would respond to medical treatments, the length of time that the child could be expected to live, and how their ability to manage would be altered as the child's disease progressed. The uncertain course of the child's illness caused enormous stress for families and contributed to increased intrusiveness on their lives. They were taken on a "roller coaster ride" over which they had little control.

In addition, the illness experience placed heavy demands on families and taxed them cognitively, emotionally, and physically. Families used the process of *navigating uncharted*

territory (see Figure 1) to manage those demands and, thus, gained some control over their lives. The extent to which families integrated the various themes within the process influenced the final outcomes. For example, while parents might have confidence in their ability to provide physical care to the child, they also needed to make sense of the situation if they were to be satisfied with the experience. Thus, the outcomes of *navigating uncharted territory* reflected the families' abilities to successfully manage the cognitive, physical, and emotional work of the illness experience. It is important to note, however, that none of the families' reactions should be considered "abnormal" or "pathological." While there was a range or continuum of reactions, all of the reactions should be viewed as "normal."

Emotions as Momentum for the Process

Strong emotions of fear, uncertainty, and grief gave momentum to the process. While these emotions were always present throughout the process, they changed in intensity over time. Fear and uncertainty were stronger at the beginning when parents were aware that something was wrong with the child. Before diagnosis, parents were terrified that the child would die. They did not know what would happen or what they could do. They lived with fear and uncertainty. Then, they learned that their "worst nightmare had come true" and the child would definitely die. However, they did not know how or when. In many cases, the fear lessened immediately after receiving a diagnosis, while the uncertainty remained stable or intensified. As parents employed strategies to manage the cognitive and emotional work, the fear and uncertainty dwindled. Meanwhile, parents were also grieving.

The first six to twelve months following the child's diagnosis were ones of intense grieving for all families. Gradually, the intensity lessened as families "got on" with their lives, but the grieving never went away. As one mother said:

You have a little period of your day where you do your little grieving kind of thing. Then you get out of it. You move on with your day and just enjoy the day.

While most families reached this point, in one family, the father's grief never lessened and he was virtually unable to function. He gave up work and was consumed by thoughts of his child's death. In another family, a mother also continued with intense grieving. Both these parents were receiving anti-depressant treatment because of their inability to "get on" with their lives.

For many families, "getting on with life" seemed to almost "just happen." Some parents had started to make adjustments in the home to accommodate the child's bed in the living room, while others were making plans for the child starting school when they realized that the intense grief was gone. Regardless, families accepted that they had to cope with their child and live with the prognosis. It was a matter of fact and they could not change it. As one mother expressed it:

Accepting that this is going to happen, no matter what you do. No matter if you get depressed, no matter if you stay happy, it's going to happen regardless of how you accept it or go through with it. So, once you've accepted that, then I think you can move on better, in my opinion.

During the time following their child's diagnosis, families experienced many losses that contributed to their grief. They lost their dreams for the child, and whatever they had planned for their lives was destroyed. One mother said:

Your dreams die. The majority of your dreams just go out the window when your child is diagnosed. Yeah, you lose your dreams. You lose your dreams for that child. You lose your dreams for what your family is going to look like.

Things like going into Zellers. I'm in Zellers and I'm doing some shopping. I'm looking around. I go to the Girls 7-14 sizes, which is her size, and I'm looking at clothes. All of a sudden it hits me that I shouldn't be doing this this way. She should be there. She should be picking out her own clothes. The realization that I would never do that with her was horrible. I ran into where the pillows and all the quilts were and stood there crying. It was just so devastating to realize that loss.

Families also accumulated many "smaller" losses. They watched as the children lost motor and verbal skills or required increasingly complex equipment. Parents had to adjust to each loss. Yet, there was seldom time to adjust to one loss before another loss occurred. The magnitude of the distress caused by what might seem, on the surface, small or insignificant losses was not apparent to all professionals. This mother ruminated:

Maybe it's - yes it is anticipatory grief. All of the little losses that I've had...not only with her being sick. Yes, you are thinking, "This is it. She is going to die." That wasn't what was bothering me. What was bothering me was the fact that she is now on oxygen....[Home care nurse] said he's done work with AIDS patients. He did some study on anticipatory grief. He was saying that one thing he noticed with people who have progressive illnesses is...in most normal people's lives you have a loss of some kind - whether it is your children growing up or what not - but you usually have time to adjust. Whereas with us, it is one after the other after the other after the other. She [child] now lost the ability to breathe on her own, she has to have oxygen, she is not going to school full-time - so there is a loss.

Another loss families experienced was for the children and the family they might have had. Since many of the NLTIs were genetic, parents often chose to have no more children once they received the child's diagnosis. In most cases, parents had been told by a geneticist that the odds of another child having the disease were one in four. These odds were too high for some families, but seemed very reasonable to others. One mother with two children said:

We wanted three [children]. We were definitely going to have another child. Then, once our daughter was diagnosed...we won't, we just could not. We don't feel it would be responsible to bring another life into this world and for them to perhaps have this disease. It is just not fair to them. It is not fair to the sibling and I couldn't go through it again. I could not.

This family was against abortion, so they would not risk another pregnancy. Other families were willing to "risk the odds" and try again. No one, however, was willing to have another child with a similar condition. One mother said:

I still want more children. I am willing to do the amnio[centesis]. I am willing to deal with the fact that yes, if it was another child with this [disease] then I would abort. It's just not fair. It's not fair to the child and it's not fair to the family to do it more than once. It's just not right. This happened for a reason. I wouldn't take the chance again. The amnio[centesis] would say definitely. There's a one in four chance, so the odds are pretty good. There's a one in four chance that yes, the child would definitely have it. A two in four chance of the child being a carrier. And a one in four chance of the child [having] no effects whatsoever. So, they're pretty good odds considering a lot of the disorders that are out there. The odds were pretty good, so I am willing to take the chance.

Some families were willing to consider adopting other children, but not until their child had died. They did not think that they could manage another child while they were focused on their ill child.

Siblings also experienced losses as the child's condition deteriorated. Their friendship and sibling relationship with the child was altered. They no longer had a playmate who could run around with them. They also often lost time alone with their parents. Many times, siblings received less attention from their parents because attention was now focused on the ill child. Finally, families experienced losses of relationships with extended family and friends as a consequence of focusing on the ill child.

Secondary precipitating events in the process sent families into unfamiliar territory once again. These subsequent events held the potential to intensify emotions. Differences in the intensity of each emotion depended on the particular precipitating event. For example, changes in the child's condition intensified fear, uncertainty, and grief. Parents were afraid that each change signified imminent death. Another event might only intensify the uncertainty. If a child was getting taller and heavier, then parents were uncertain about the availability of different equipment to help them. They were again in unfamiliar territory, but this type of event was not accompanied by increased fear and grief.

Grieving was ongoing in all families, but some times were worse than others. Other events that triggered a resurgence of grief included birthdays, Christmas, seeing other children of the same age starting school, or just watching other children. In one family, parents were emotionally drained after their child appeared for the second time on a television telethon. They then saw their child "through the eyes of someone else" and were confronted with how many skills the child had lost in a couple of years. The parents described this realization as "devastating." Families felt as if they were on a "roller coaster" of grieving that would be with them forever.

Physical Impact on Families

Families expended considerable energy as they identified and implemented strategies to handle the cognitive and physical work while attending to the experiential nature of the illness

experience. Life was a never ending juggling act that involved complex managerial tasks.

Amongst other activities, parents investigated available services, lobbied for and co-ordinated those services, prepared restricted diets, attended medical and therapy appointments, and worked hard to maintain or improve the child's health status. At the same time, families attempted to complete the usual activities of daily living.

The constant, unrelenting nature of the 24-hours-a-day, 7-days-a-week care required by the child took a physical toll on parents. Many suffered from exhaustion, injuries, migraine headaches, anemia, or hives. Parents reported that the months and years of not sleeping were the "worst times of our lives." They were on call 24 hours a day. They were often up at night for hours and became exhausted. Yet, they needed to function at work the next day or continue caring for the child while carrying on with daily living. One father stated that he took naps during the day so he could keep going. He felt lucky that his employer supported him by allowing flexibility in his schedule. Without such support, he could not continue working. Other parents felt that they had no choice but to quit work. They could not do "two jobs."

Families who had a nurse at night, or who scheduled the night time hours so that each parent could sleep for four hours and be on duty for four hours, managed fairly well. However, these strategies were often not put in place until after many months or even years of sleepless nights. Where possible, parents tried to get to bed early, but this left them little, if any time to spend together as husband and wife. While some parents used sleeping pills at different times, they disliked the side effects that left them feeling incapable of caring for their child.

As children deteriorated, they became unable to move or walk and they needed to be lifted and carried everywhere. Yet, as time passed, they were growing and becoming heavier. Parents often needed to lift a child of 30-40 pounds plus a wheelchair that weighed another 30 pounds. These children lost muscle control. They were a dead weight when being lifted in and out of the bath, and could not hang on to their parents. Many parents complained of sore backs.

The cost of mechanical bath lifts was often prohibitive. In addition, such lifts were large and not designed for use in a small home, so there was no space for them. Some professionals provided useful advice to parents to alleviate or prevent injuries, such as removing glass shower doors or using a height adjustable bed for the child. Not all advice was helpful. One set of parents asked their social worker for help in obtaining special bath equipment that would reduce the stress on their already injured backs. They were told to put a towel on the floor in the kitchen and bathe the child in a plastic bath on the floor. Yet, neither parent could bend down low because of their back injuries. Inappropriate advice such as this caused anger and frustration for parents.

Emotional Impact on Families

In addition to the physical toll, life with a child who had an NLT I caused parents extreme emotional distress, particularly in the early stages of the process. Parents were often scared, especially when the child was first ill and whenever the child's condition deteriorated. All were emphatic that they would never knowingly put themselves in a similar situation. While some parents reported that they might choose to risk another pregnancy, they would only do so if a diagnostic fetal test was available for the NLT I. They would not give birth to another affected child. It was too hard on everyone involved, including the child, siblings, parents, extended family, and friends. One father said:

A part of us is dying with our son. A part of our emotions is going with him. We don't know what is going to happen. We don't even know what state it is going to leave us in at the end. It will be almost like a sponge that has been wrung out.

Many parents were deeply sad, cried a lot, experienced anxiety attacks, or felt guilty about a number of issues. Because many of the neurodegenerative diseases were genetic, some parents felt responsible for causing the child's illness:

I think in my situation it's because I feel a lot of guilt, because it's a genetic disease and it just happened to come from me. So sometimes I find myself thinking I OWE him this. Then other times I say, "Well, it's not that I owe him. This is what I SHOULD be doing."

Some of the treatments that children received were experimental. Parents questioned whether they had made the right decision to put their child through a treatment that perhaps caused pain, had negative, long-term consequences, or that prolonged quantity of life without allowing quality of life. This mother of two affected children recognized that the experimental treatment given to one child had slowed the progression of the disease compared with her other child's condition, yet she asked:

"What have we done? What quality of life have we locked her into?" That's what I have to deal with now. I messed around with it. I let her go through with the bone marrow transplant. "But what have I done to her? Might it have been better for her to have gone through a graceful ending?" That part worries me now because I have prolonged her life. I hope that I will outlive her, because if I don't then I shall really feel as though I've done an injustice by doing what I did. But at the time, you do what you think is best.

Parents also felt guilty for accepting help, whether it was in the form of money, equipment, or respite care. As one father said:

Yes, it is hard because you don't feel that you are needing it in a lot of ways. There is someone worse off than you are that needs it before you do. So there is always that in the back of your mind, "Well, maybe I don't really need it and someone else really could use it more than I could." So, yeah, you hesitate, but it gets down to a point that [if] you don't ask, you are in a worse state than you are now. So, you feel you have to because it isn't for you, it's for your child.

This father's guilt was alleviated when he attended a course about chronically ill children and realized that "you cannot do it all by yourself." He also learned to ask for help "because the child deserves all the help that is available."

Thinking about the future when the child had died also caused feelings of guilt:

I used to feel guilty whenever I would think about what it would be like afterwards [when the child has died]. I would almost feel giddy because I kept thinking of all this freedom I'd have. Then I'd feel real guilty about it.

Most parents spent so much time and energy thinking about their child that they were unable to concentrate on anything else. Even reading a book became almost impossible for many people. Two fathers gave up work because they were unable to concentrate on the job at hand. Instead, their thoughts were filled with their child. As one father explained:

If you tried to concentrate, you would be wasting your time. You would just be wasting [time] and spinning your wheels. You would have to go back and re-read it again because you forgot, because it wasn't stored anywhere...you are too tired to really store it. It is like cramming for a study session. After four days and you've still got two more days of it to do, and on about the fifth day you are going, "Forget it. I won't get anything else in there, it's full."

Another father said that he was psychologically affected and had lost many of the analytical skills he had possessed before his child became ill. He added:

You lose your ability to think and are living a "treadmill existence." It is too stressful to think about something new, even if it's a better way of doing something than what you already do. It is much easier to continue as you are. Anything new becomes another pressure.

Others parents were clinically depressed. A couple of parents had contemplated or actually attempted suicide. One father reported that he still welcomed the idea of death. Some parents were angry or resentful about the unfairness of the situation. They became angry with health care professionals or with God. Others felt out of control and helpless. As one mother said, "When they've [one's children] got something and there's nothing that you can do to make them better, it's the most awful thing. You just can't describe that. It's a hopeless feeling."

Many parents used anti-depressants, alcohol, cigarettes, or illicit drugs such as marijuana to block out the pain they were suffering. Cigarettes also relieved stress for some parents. While counselling could be helpful, especially for mothers, many could not afford the cost of a counsellor. Psychologist services were not covered by government programs. Emotional distress changed and often lessened as parents became more familiar with the illness, its treatment, and prognosis. However, changes in the child's health, such as an acute chest infection, inevitably produced increased distress as families were thrown into unfamiliar territory.

While there were few siblings in these families, both parents and siblings themselves identified a number of emotional impacts. Siblings were sad that the child was dying and that they were losing a playmate and sibling. They were also angry that the child was dying, and angry or upset that their parents and others focused so much attention on the child that the

siblings received less attention than they wanted. The increased number of people in the home was stressful, although sometimes it allowed siblings to "get away with more" because they could go to the professionals when parents disciplined them. One sibling feared that her parents would separate. According to this sibling's mother, "She really hasn't adjusted. She gets upset that people talk about her sister and not to her. You saw how she looked when you came to the door" [sibling looked crestfallen and somewhat annoyed].

On the positive side, a couple of siblings were quite self-sufficient and confident because of, in their parents' opinion, their upbringing. They were adaptable to changing circumstances. One mother commented that the sibling was "very empathetic." She believed this would "stand the sibling in good stead the rest of her life." While siblings were sometimes demanding, they were normally viewed by parents and others as less demanding than other children of their age. In addition, some siblings were capable of reasoning abilities that would be expected of an older child.

Parents did not expect others to fully understand their experience. As one father said "Something like this, most people do not experience in their lifetime. You might see it from a distance, but unless you actually go through it, you never know what it is really like." However, they hoped fervently that extended family, friends, professionals would learn as much as they could about the family's situation. Thus, they could become empathetic and could provide the type of support that families needed. Parents did not want others to experience a similar situation or to feel the emotions that parents felt. However, they wanted others to gain an appreciation of how difficult life is for families. They suggested that recognizing the family's inability to do anything on the spur of the moment would be very helpful. One father wished that other people would:

Take us into account first if they start making plans, rather than make the plans and then see if we can fit into those plans....Maybe they should find out what fits our schedule or situation first and then make plans around that.

Families had varying success on their journeys, especially related to their ability to manage the emotional work. While all families underwent the process of *navigating uncharted territory*, because they had no choice and could not change their child's illness, some parents also underwent a transformation that resulted in personal growth. Those parents who made peace with their emotions, accepted the situation, and created positive meanings out of the experience felt more in control of their lives, were not overwhelmed by emotions such as anxiety and depression, and believed that they had gained from the experience.

Navigating Uncharted Territory: A Basic Social Process

The basic social process of *navigating uncharted territory* was comprised of four main dimensions: *entering unfamiliar territory*; *shifting priorities*; *creating meaning*; and *holding the fort*. While they are presented sequentially for ease of explanation, these dimensions were overlapping and iterative. For example, families continually created and recreated meaning in their situations. In addition, families entered unfamiliar territory at the beginning of the process. However, whenever they were faced with a new situation, such as a deterioration in the child's physical condition, they often again entered unfamiliar territory. The behaviours of families were related to the stage of the child's illness. Changes in parental strategies were closely linked to pivotal events in the illness trajectory. Each of the dimensions in this process will be explicated in the following section. The various management strategies used in each dimension will be identified, as well as the consequences of the strategies.

Entering Unfamiliar Territory

Families entered unfamiliar territory whenever they encountered a new situation. Entering unfamiliar territory in the context of a child with an NLTI was characterized by strong emotional reactions to a perceived threat to the child's health or life. Fear and uncertainty were evident as families first faced the possibility that the child was "abnormal" in some way. Once a

diagnosis was made, they also grieved for their child. Their fear and uncertainty pushed parents to look for answers to questions about the child's illness, treatment, prognosis. These emotions drove parents to find information and to learn how to manage the situation. At the same time, they needed to deal with their grief. The process of *navigating uncharted territory* was initiated by a precipitating event, such as a parent recognizing an unusual behaviour in the child. Subsequent events, such as the child developing an infection, continued the process and sent families into unfamiliar territory over and over again.

Initial Precipitating Event

The initial precipitating event that started the process of *navigating uncharted territory* invoked visceral reactions in parents who were caught up in an overwhelming experience. The fear, agony, anguish, and terror they felt because their child was sick overpowered them. Yet, these emotions were not always obvious to friends, extended family, or professionals. One mother said:

During that period of time [prior to diagnosis], I think I was just - You just go on. But, you are just so very frantically scared. I remember somebody asked me what that was like and I said, "It is [like] you're screaming in your head all of the time because you are so terrified." But, you just - On the outside you come across as you are okay....For me, I was just screaming inside for months and months and months just out of sheer fear.

Families first entered unfamiliar territory when they noticed that something was wrong with their child. Changes in behaviour or motor skills, such as fine tremors or falling over for no reason, were often very subtle. Becoming aware that their child was "not normal" was often an insidious process that happened over a period of time. Mothers were frequently the first to suspect that there was a problem long before the child was diagnosed:

She was clenching her fists. She'd come to me and she'd say, "Mum. Look at my hands." She'd open them up and they'd all be sweaty. I'd tell her to try not to keep her fists clenched, try to keep her hands open. That the clenching was causing the sweating. She was always a very outgoing child. Had hundreds of friends. Just loved to be the centre of attention. She began to withdraw. She pulled away from her friends. She'd sit down more. Our neighbours had a little baby. She'd play with the little baby because she could play with the little baby sitting down. I noticed these things and I couldn't understand

what was going on. I knew the Christmas before she was diagnosed. I knew something was going on.

At first, other members of the family did not notice the changes. Mothers often felt alone because they were the first to notice a problem. One mother said:

I had this terrible fear. When she was 8 months old, I remember crying. Alan [husband] came home from work. He said, "What's the matter?" I said, "I don't know what's wrong, but I don't think Amanda's ever going to walk." He said to me, "You are just a first time mum and don't worry about it."

As the changes became more obvious, other members of the family started to notice them too. Sometimes it took a few months before this occurred. Families were scared, worried about the child, and fearful of what was happening. One mother remembered, "Even before she was diagnosed we knew. I had this terrible fear. We lived with this fear because you would see her degenerating or losing skills."

Diagnostic Process

The diagnostic process was usually challenging for parents. It often involved a lengthy and frustrating search for answers. Mothers typically instigated the detective work while professionals did not appear concerned. In many cases, professionals minimized or ignored symptoms and complaints. Parents often found it difficult to attract enough attention to even get diagnostic tests done. They needed to be persistent and had to work hard at convincing others that the child needed further investigation:

I'd call her [child] and sometimes she wouldn't hear. Alan [husband] had problems with his ears when he was a kid, so I would go to the doctor and I'd say, "I think she has got an ear infection, could you please check her ears?" Well, he would check her ears and there was nothing wrong. There were times where we would be driving in the summer time and it was like she had fainted. I'd never seen a seizure in my whole life so I had no idea what was going on and I went into his office. In fact, I was driving down the road and her eyes rolled up and she fell over and then she sat up again and her eyes rolled up and she fell over again and I drove directly to the doctor's office. I said, "I don't know what's wrong, but she's kind of done this thing." He is questioning me and he goes, "It is just heat stroke, so make sure you get lots of fluids into her." So, fine, it didn't happen again and the doctor just said, "You are an anxious mum. Don't worry, every child develops differently." She wasn't speaking all that much [either].

Perseverance paid off for some families. In others, it took a crisis, such as a major loss of motor abilities, before professionals listened to families and were also convinced that the child was ill. A serendipitous change in physicians was helpful for one family. The new physician in the practice acknowledged the mother's fears and acted on them because, as he explained to the mother, "In my experience, if a mum or dad thinks that there is something wrong, I go with that."

Once professionals acknowledged that there was something amiss with the child, they started looking for a diagnosis. Families often spent weeks, months, even years trying to find out what was wrong with their child. Some children spent half their lives in hospital during this time. Hospital visits were numerous and families were always running to appointments:

Appointments for this and appointments for that. In and out of hospital. That was a biggy. We spent a lot of time rotating between the hospital and at home in the first couple of years. Six months, it seemed like, a year, was used up. Hospital visits, so that was there and then back home and then back there and two weeks in and three weeks out and two weeks in and four weeks out. On and on it goes....But I mean, we used to be going to the hospital all the time.

Parents spent as much time as possible with the child during hospitalizations. Mothers were often the ones who attended all of the appointments. Many parents took time off of work so that they could support one another. They wanted to ensure that "everything was working out the way it should be." Parents needed to feel they were "actually on top of everything."

Children underwent "every conceivable test they could do." Nonetheless, the long process of diagnostic testing was often a frustrating quest that did not guarantee a definite answer. Inaccurate or incomplete information was sometimes given and children were misdiagnosed at times:

They came up with that she was developmentally delayed: no seizures. But within six months, she started to seize. Then they diagnosed her with epilepsy, and then she just kind of went down and down and down after that.

Sometimes professionals suspected the particular NLTI, but it took a long time for a final diagnosis. Professionals wanted to be positive of the diagnosis before they told families that their

child was dying. In addition, testing facilities were usually not available in Canada and the tests had to be done overseas.

Parents felt helpless and out of control as they searched for a diagnosis. "It was really hard in the beginning, like when they were diagnosing him. You don't know what to do." They were terrified because they could see their child losing abilities, yet no explanation could be found.

Once a diagnosis was made, professionals then faced the challenge of telling the parents. All parents had profound and vivid memories about how they were first told the diagnosis. While the diagnosis was expected by some parents, it was a complete surprise to others. Each parent was shocked, disturbed, and in great pain. The timing and setting of the diagnostic event held major significance for parents. Most parents appreciated the attempts made by some professionals to break the news gently:

The doctors were very good in that they tried to make it - I think they knew all along.... You could tell from the symptoms, from his features and everything else, that this was probably a metabolic disease. But they had to confirm it with tests.... Almost every week we would have a meeting. We would sit down and they'd come back and say, "Well, we've done this and this test, and the results aren't very pleasing. I'm afraid your son is going to lose his eyesight."

Other parents were shocked by the manner in which they received the diagnosis. They were overwhelmed by the diagnosis, and by their new perception of their child as a dying child. They felt a sense of urgency and anxiety that often went unrecognized by professionals. Some health care professionals seemed to minimize or disregard the existential significance of a terminal diagnosis. One mother received the diagnosis during a chance encounter with the child's physician in a parking lot. She had little time to assimilate the information or to ask questions before the physician left her alone. The mother had no recollection of her drive home with her two children because she was in deep shock. Another family was told the diagnosis by telephone in a brief interaction that resulted in an angry physician refusing to discuss the meaning of the

diagnosis. One father shared his feelings about how he was told his daughter had an incurable illness:

All the neurological doctors at the hospital should have a frozen boot shoved right up their ass for the way they told us about Sarah. I will never forgive them for that. I was sitting there. They've had their battery of tests. Some hobnob comes in, says, "You're Mr. A? Let's have a chat." Pulls me into a semi-quasi broom closet/lunch room. Says, "Okay. She's got this disease. It's going to go like this and it's not very good. Okay, I've got rounds, see you later."...I said just a moment. He said, "Yes?" I said, what do you mean? He said, "Well, it's pretty" - I don't know if he said it's pretty self-explanatory, but this is how it felt. He said, "Well, she's got this, this and this." I said, hold on. Whose going to tell my wife this? Because I don't have, I'm not that deep on the realm of what you've just said. I know my wife will have a lot of questions, so who is going to? From what you've told me, this is not good and I will not be prepared to answer all the questions for my wife. I don't know what to do. I don't know what he said, but he just sort of walked off. Then, one of the residents said to me, "Mr. A., I'll come and I'll be with you there."

Parents expected sympathy from the professionals. But more importantly, they expected honesty and they did not always get that. For example, one family was told that the child had an NLTI, but the parents were not told what that meant or even which particular disease the child had. Professional honesty facilitated discussion about the meaning that was embedded in the diagnosis. For many parents, the most important meaning was bringing closure to a long and frustrating diagnostic process. In many cases, a definitive diagnosis validated the parents' suspicions and complaints. The time of diagnosis was also a critical turning point in families' lives. It signified the end of a part of their previous lives. It was difficult for families to hear the diagnosis. Yet, they often expressed relief that the cause of their child's problems had finally been identified. They were frightened and they started grieving, but at least they now knew what they were facing and they could "go on and start to cope." One mother described it this way:

So he [physician] proceeded to tell me and it was just like this major calming. Just the fear. Yes, there was still fear, but not that fear of the unknown or imagining how bad it would be. It was the worst that you could possibly hear - that your child is going to die from this - but somehow it just, it was just this sense of calm and a big relief to finally know. He had to get up and leave the room, because he was just very overwhelmed. He was starting to cry. I thought he was going to leave. I chased after him in the hallway. He is standing out there like this [head down and upset]. I said, "Thank you, thank you, thank you for telling me." I think back now and I think, here I am thanking this doctor, and I'm

telling him, "Thank you for telling me that my child is going to die." Of all things. Then he's crying and I'm crying and the nurses are crying. But it was a major sense of relief to at least have some idea of what we would be facing.

For many families, the diagnosis marked a change in their relationships with health care professionals. They found that professionals lost interest in the child once a diagnosis had been made and/or no cure was possible. Some parents were also disillusioned that the diagnosis provided little useful direction and little help from health care professionals. Most families felt supported by staff while the child was receiving tests and treatments. Once no further treatment could be given and the child was receiving strictly palliative, not "curative" care, then some families felt alone, scared, and "let down" by the system. Contact with hospitals became sporadic, and often only occurred at scheduled yearly visits or when the child had a crisis. This mother and father described their experience:

Father: When people like us end up in your world, don't leave us alone. Just don't shove us out of the door and leave us alone.

Mother: Yeah, that's what everybody's done.

Father: Because THAT has been the worst.

Mother: Because Sam and I were so involved in everything. Everything that the doctors and nurses did to Ian, Sam and I needed to know why, what, where, when, how - everything. We had meetings every Friday because Sam and I became very, very frustrated with the whole system and said, "Okay, you're talking team. We're a part of this team." So finally, they started to listen to Sam and I, really listen. Sometimes I would just babble on about his [child's] bowel movements, because that to me, that was my job....Then, once we left, that was it. That was IT. It was like they were throwing us to the wolves.

Seeking Information

Parents entered unfamiliar territory when their child was diagnosed. Moreover, they were often presented with new situations or tasks throughout their child's illness. Whenever parents entered unfamiliar territory, they sought information to help them manage the new territory. The impact on parents of having received a diagnosis was an immediate and urgent need for information. In many cases, mothers were more insistent than fathers on seeking answers to why

the child was ill and what could be done. At first, parents wanted to change the course of the illness. Therefore, they looked for information about the disease and its treatment. Many parents learned how to access and use medical libraries and the internet. They taught themselves how to read and evaluate research reports. They also contacted international experts on the child's condition and attended conferences to learn as much as they could. Families followed every lead they could get in their hunt for information. They often gained the most information from lay support groups and other families with a child who had a similar condition.

Over time, most parents realized that their child would not be cured. They no longer searched for a treatment, but worked at making the child as comfortable as possible. Parents needed honest and sufficient information so that they could learn how to cope with the physical, cognitive, and emotional demands placed on them by the child's illness. This father said about himself and his wife:

We tried to educate ourselves with as much information as we could. We spoke to the clinical nurse and saw a video on tube feeding - how the operation is done, what it meant, the care and the upkeep and all that. We actually asked to see a child that had a tube in him at that hospital - that helped. There again, the more information you have, the better. Some of it may not be pleasant, but at least - if you have somebody to hold your hand and walk through it, that helps a lot.

Parents' need for information was often pressing, yet unmet. Looking for information was often stressful for parents, because they had little time in which to do it and also because they had no guidance. Many parents felt as if they were "running all over the place....A lot of [learning] has been trial and error....The onus is always on the parents to go out and find it." They appreciated receiving early direction and anticipatory information and guidance from professionals. Some families were extremely resourceful and creative, but others were less skilled at meeting their information needs. Acquiring a sophisticated knowledge of the child's illness and its management required an extensive time commitment when free time was usually limited. It also required some dedication to learning and access to knowledge resources.

An intensive search for information about the illness was often frustrating and unhelpful because information was either non-existent or difficult to access. In addition, once parents learned about the genetic nature of many of these NLTIs, they faced the difficult issue of genetic testing for siblings. The siblings in two families were tested. Parents waited anxiously for the results which again took a long time. One family was delighted to learn that the sibling was unaffected. The other family discovered that another one of their children was affected, though the third child was "normal." Knowledge about genetics also had implications for conceiving other children.

Information needs changed over time in response to issues or difficulties that arose during the course of the child's illness. The child's physical condition necessitated that parents learn many new things. For example, as the children grew, they were often unable to wear normal clothing because their limbs were twisted, or parents found it too difficult to struggle with buttons. Parents then searched for information about companies that made "special clothes for special kids." Most children acquired multiple pieces of equipment, such as oxygen, suction machines, and feeding pumps, all of which families were required to use by themselves at home. Families learned how to clean gastrostomy tubes, change naso-gastric tubes, and give postural drainage. Parents tried to educate themselves with as much information as possible so they could provide the most effective care. In many instances, professionals in the hospital provided training, but sometimes families felt inadequately prepared before the professionals wanted to send them home. Parents had to be assertive and insist on receiving sufficient instructions. Although families were often afraid of the unknown, they were willing to learn just about anything so they could keep their child at home and still provide excellent care.

Families also needed to learn about the services that were available to them at home. Government programs that provided the majority of services offered some free professional care, medications, and equipment to families in their homes. Families learned about programs in a

number of ways, such as through the hospital or an organization for the disabled. However, it was often many months after the child's diagnosis that families learned about programs. In the meantime, they paid for everything by themselves. Government programs eventually provided some assistance to all families. But, they never totally covered the family's costs and parents had to continually ask what would be covered. As one mother said, "Most people don't know what the program allows or what it covers."

Equipment such as a wheelchair ramp or lift were not covered by government programs. Equipment often cost thousands of dollars that families could not afford. Parents had to seek funding on their own from other agencies and charities such as the Variety Club. Many times, families had no idea what funding and agencies existed. They spent extensive time and effort both learning about the agencies and writing to them for funds. Many times they were unsuccessful in obtaining funds because the demand outstripped the availability of money.

Other important services that families needed to learn about included local pharmacies and medical supply stores. They quickly learned which pharmacy was open at midnight in case they needed to rush out for medication because their child had a fever. They needed to know what days a medical store delivered and how quickly an order would be filled so they could plan when to order supplies. Local centres for the developmentally delayed also provided assistance to families, but each family needed to learn what was available in their community.

As well as learning how to manage the physical and cognitive work involved in caring for their child, families had to manage the raw emotions that arose from their situation. Many parents needed outside assistance, at least in the beginning, to help them deal with their emotional responses. A counsellor or psychologist was the most effective professional for those family members who wanted this kind of support. They wanted someone "you can go and unload to, who can often come up with some ideas. Somebody who understands." One mother of two ill children noted:

In the beginning, I saw an awful lot of psychologists. I don't have any hang-ups about it. They're professionals and they have skills that I don't have. So, if they've got the skills then help me. We were seeing an enormous number. Well, I was seeing an enormous number. I saw one for myself. Tom [husband] and I saw one to help us through. I saw one at Children's [hospital] for Nancy [child2]. I saw one for Sarah [child1]. Then there was one at school that was talking to us. I would talk to as many people as I could to get as much help as I could. I think that that helped me get through and gave me the strength to help them get through.

Even though psychologist visits were often very helpful, some parents were forced to discontinue them because they could not afford the fees. They were often angry and bitter that the health care system would not help them learn how to deal with very painful emotions. Those parents struggled with managing the emotional work. They did not have an outlet for their emotions. Nor did they have someone who could suggest ways of managing their emotions.

Counselling was not helpful for every person. Some parents needed more than just an outlet for their emotions. They wanted a counsellor to provide concrete ideas and solutions to the parent's perceived problem. These parents expected solutions. They were disillusioned and disappointed when no solutions were forthcoming. One father said:

We've been to the psychologist. But my feeling is he doesn't address the issues that are at hand. So why should I go? He was not working. He listened [emphasis] to her [wife] and he listened [emphasis] to me, but how long are we just going to listen?... Sometimes it's just nice to vent. People listen and it does make you feel good. But I want solutions.... What it did was it set me up for expectations. This is the most important thing - it set me up for expectations. When they didn't arrive, that made the fall worse.

In addition, some parents preferred to keep their emotions to themselves and did not want to discuss them with others, including psychologists.

As time went on, parents made contacts and gained skills that increased their confidence in their ability to search out information. They increased their advocacy and negotiation skills. Parents usually knew where to go for information. They learned who to ask, as well as how to ask for information in the most effective way. Most parents eventually became very efficient at seeking information. However, it was a very time consuming process that depleted their limited energy.

Sharing Information

Parents sought information whenever they were in unfamiliar territory. Sharing information was another strategy some parents used when they were in unfamiliar territory. Once families obtained information and became more familiar with the new territory, some of them wanted to share the information with others. They did not want other families to have to go through the same difficulties that they had faced. Parents spoke with other parents individually and at support groups. They passed on the benefit of their experience. At least one parent wanted to write a book for other families about her family's experience. Another family was considering developing a resource manual for families when they first receive the child's diagnosis.

Not only did families learn from professionals, but they also taught professionals. They spoke with professionals to raise their awareness of "what it's like to be in our shoes." They also shared the information they had obtained so that professionals could help other families. Some families perceived, however, that the professionals did not use the information available to them:

We photocopied everything that we had and brought it into the hospital. Where that went to, of course I don't know. We just felt that they should have certain information as well, as we had the same. But where it went to, I don't know. I don't know. I don't even know if they even read it to be honest.

Parents noted the importance of sharing information with the broader society, friends, extended family, and policy makers in order to help them understand what these families are dealing with. They also wanted to raise awareness of how disabled people could and should be incorporated into societal planning, such as ensuring that buildings are wheelchair accessible.

In summary, families entered unfamiliar territory on many occasions throughout their experience. They sought information to assist them in understanding the situation and to allow them to manage the physical, cognitive, and emotional work accompanying the experience. Information needs changed over time in response to issues or difficulties that arose during the course of the child's illness. For many families, accessing information was a frustrating and

complex process that required a lot of time and energy when these resources were already in short supply. Few parents received guidance from professionals in their search for information. Once parents obtained information and became more familiar with the new territory, they shared the information with other families and professionals. They did not want other families to have to go through the same difficulties that they had faced. Finally, information provided a basis for some of the strategies parents used in the other dimensions of *navigating uncharted territory*.

Shifting Priorities

The second major dimension of *navigating uncharted territory* involved a shift in priorities within families. While priorities shifted when the child was diagnosed, shifts also occurred following each crisis or whenever families entered unfamiliar territory. Once the child's diagnosis had been made, parents often reported feeling relieved and more calm. However, they were still overwhelmed by the situation. Parents were filled with uncertainty. They were grieving. They faced the dissolution of their hopes and dreams. Their view of the future was limited temporally and qualitatively. Yet, at the same time they felt as if they were running and trying to catch up with the situation. Parents needed to slow down in order to gain some control. Priorities changed within families. Family members focused on the child, even to the point of excluding other members of the immediate family, instead of the priorities they had previously selected for themselves or their families. They put their own lives on hold and often withdrew from other people. Some parents accepted their child's disability and found satisfactory ways to live in the world of disability. Others struggled with wanting a "normal" life and remained unhappy with their situation.

Family priorities changed and evolved over time as families gradually began to manage the situation. In families where the child had been ill for a number of years, some family members began to think a little more about themselves and the family unit, excluding the ill child. They recognized that the family was in danger of falling apart because of the focus on the

child. They were able to think a little about what life would be like after the child had died.

Although the child remained a constant focus in their lives, they were able to use respite time to get away from the house for a while. They could start concentrating on other aspects of the family's life. Attention slowly shifted away from the child and these families began to take some time for themselves without including the ill child. Some families went on short vacations without the child. If the child's condition worsened at any time, then attention was immediately focused back on the child:

Okay, our whole life centres around Amanda [ill child]....I think when you have children your whole life changes, but when Amanda was about 18 months it was another change again and so then everything started to focus on her. I was pregnant with Janie and Janie was born. You always assimilate the second child a lot easier, but it was just, Amanda was just the centre. Like we were working so hard to try and figure out what it was. We were working hard with Amanda, and Janie just kind of got assimilated into that. We are very conscious about keeping her involved and stuff, but I mean everything has to revolve around Amanda - her feeding schedules, everything. That is the first thing that we do and then we schedule time for Janie. We've learned, I think over time, to take time for ourselves, or to do things with Janie.

None of the families perceived the child as a burden. However, a family life that was wholly focused on one child was difficult for every family. In some families, the constant focus on the child was detrimental to other family members and to the family unit. In a few families, the parents had discussed placing their child in a care facility, such as a group home or long-term hospital, in order to keep the rest of the family intact. Unfortunately, there were very few, if any, suitable facilities for children and so there were very few options available to families. Any discussion about placement caused anguish for parents, but sometimes the family saw it as the only long-term alternative if they wanted their family to survive. One mother said:

We've thought of it a few times. I've really thought of it, because it's just so stressful and so chaotic that there's no [peace at home]. I mean, we love Alexa dearly and I know for me it would probably kill me if I did that....But sometimes I wonder if it's better for her, better for our family.

And the father agreed:

We know that we're going to have to give her up into a home when she gets older and we get older. I mean, we've got to have some life, or whatever's left by the time these kids grow up and are out of the house. Are we going to tie ourselves down till we die? Because that's basically what's going to happen if we do. And we don't know how long Alexa's going to live. Things are getting better with her illness. They're understanding it a bit more, but there's still a lot of them [children] dying in their teens.

Parents used various strategies as they shifted priorities in their lives. These strategies - *going into slow motion, focusing on child, putting life on hold, getting rid of excess baggage, and picking up the badge of disability* - enabled families to manage the situation and are described in the following section.

Going into Slow Motion

Families talked about slowing down the pace of their lives so that they could manage their situation. *Going into slow motion* involved a deliberate intention on the part of families to stop moving as quickly as they had been doing before the child became ill. One father explained:

Yes, that is very, very important, because we are all caught up in our own paces. I mean going to work, rushing, just zooming around, getting shopping done and everything. Which is fine, but when you have a crisis situation like this, you can't afford to carry on at that pace.... We used to zoom around at 100 km/hour. The whole day, it was zoom, zoom, zoom, everywhere. Now we've slowed ourselves right down to 10 km/hour. But, it is knowing that, that you are capable of doing it, and being able to do it. You have to recognize in yourself, that no, if you are going to carry on at 100 km/hour, you're not going to survive for three days.... We might be moving at almost a snail's pace but it doesn't matter.... They often talk about a human race. For us, this is a race. This is a marathon for us and for us the objective is to finish the race, to finish the marathon. It doesn't matter that we are not speeding along with everybody else at 100 km. If we are going 10 km, it doesn't matter. We will get there and we will go at our pace and we will survive it because we know enough about Mike's condition. We know that we have to go very, very slow, go at his pace and that is the most important thing. I prefer going at 10 km/hour, because I know at the slow and steady pace, I am able to survive, and for me, that is a bottom line.

Going into slow motion also allowed parents to savour every moment with their child:

I make every minute last as long as I can. I've also kept a journal from the time this happened. Every day I write in this journal. It's Ian's journal actually. I say all the things that he does on a daily basis. Whether it's something that someone would think would be stupid - of course, it's not to me. That's the way time has changed. I'm no longer in a hurry. I can do everything at a slow pace. I don't rush Ian, EVER. I get him up at 6.30 in the morning to be on the bus at 8 o'clock. That allows us time to eat, and to play around, and to get changed. It's very slow, I'm at a slower pace. Time with him is... I get scared

when I go into from one month to the next, because it's "agh, a month is gone." Even though I'm at this really slow pace, I feel sometimes that I'm in slow motion. But it just means that time is so valuable to me. From 8 o'clock in the morning till 8 o'clock at night means so much to me, the time that I get to spend with Ian.

In addition, slowing down was important in dealing with the child's condition. Changes were not tolerated well by these children. In most cases, the children required routine and order in their lives. Otherwise, they became emotionally and physically upset. Families took "everything very easy, very exploratory" when doing anything with the child. For example, parents could not simply increase the amount of fluid the child received through a tube feeding. They were aware that even a few extra millimetres of liquid could overload a child with a gastric reflux and could cause the child to aspirate. So, they increased liquids very slowly and carefully. Parents were always conscious that they could not move too fast. They were prepared to "stop and back off" at the slightest hint of any problems.

When families went into slow motion, it affected their ability to learn new things or to process information. As one father said, "If it takes a normal person one day to do something, it will take us two or three days." It then became important for professionals to slow down as well to match the slower pace of families. For example, when professionals offered new ideas or suggestions to families, such as offers of personnel or mechanical aids that seemed like a good idea from the professional's point of view, families saw it as another pressure. It was adding another stress into their life because it was something else to think about. Families needed time to work through new ideas or suggestions until they finally understood and it was no longer stressful. However, professionals and others were frequently unaware that parents required more time than usual. Families suggested that professionals needed to reduce their time and their speed to allow the families time to accept new things and to sort them out. Moreover, families believed that professionals should acknowledge that parents needed extra time and they should give parents time to think about things.

At the same time, parents acknowledged that they were often so immersed in a situation that they could not see alternatives. Parents were often "too stressed to think clearly." They needed someone else to point out when help was available and required. While parents appreciated suggestions from professionals, they also recognized that professionals could not always tell when the right time might be to give advice. They suggested that professionals should offer options and make suggestions on an ongoing basis. Thus, professionals could raise possibilities for the future as changes occurred and still allow families to take the time they needed.

To summarize, families went into slow motion because they needed to step back from the frenetic pace of diagnosis, treatment, and everyday life in order to survive. When parents slowed down their pace, they could deal with the issues that required their attention. They could also enjoy each precious moment with their dying child. *Going into slow motion* affected parents' abilities to assimilate information and make decisions. Yet, professionals were often unaware that parents needed more time. In addition, *going into slow motion* allowed parents the time and space to focus on the ill child.

Focusing on Child

All families focused the vast majority of their time and energies on the ill child, especially in the first few months or year after the diagnosis. As one father said, "You put your child ahead of you and all of your wishes all of the time." Focusing on the ill child meant that families were unable to continue their previous lifestyles. They were always dependent on what the child's condition was like and if someone was available to look after the child. Members of the family had little time to be alone and have what one family called "private time," a time of solitude to enjoy some peace and quiet or to do something they enjoyed doing such as gardening or reading a book.

In families with two affected children, family members had even less time for each other. One ill child took up much of the parents' time and energy, but two children more than doubled the work. Parents often did not realize the extent of their work until they had an opportunity to spend time with only one child:

If we want to take one of them, then I still have to get someone specialized in to look after the other one. I think that's an impact of having two high needs [children] which I think a lot of people don't realize. When we get rid of just the one and we have just one to handle, it's so much easier. You don't realize how much you're doing with two of them until you remove one from the scenario.

Unless two adults were present, one child had to wait until the other one was dealt with. When the child was fairly stable, this was not a huge problem. If a child's condition deteriorated, however, an added caregiver became imperative. For example, one father gave up coaching a sports team in the evening because he needed to be home.

Parents wanted to give the child as many experiences as possible in the limited time remaining. They knew that the child could not live life to the fullest extent. However, parents wanted to maximize the child's quality of life by providing many different experiences, such as "swimming in a lake or rolling in the grass." One mother said, "If I can make their life happy and get them some experiences while they can experience them and enjoy them, that's what I want to do for them. That's all I can do for them." Parents worked hard at involving the child in activities, so the child "did not have a boring life." For example, the child's attendance at school was important to parents because it offered exposure to a variety of experiences. Whatever length of life the child might have, parents wanted to "make the best of it."

Parents also gave the child as much as possible so the parents would have fewer regrets about the care they provided. One father explained:

When you have a child like this, it is important to do the best you can and have no regrets. That is very, very important. Because if you have a regret, what happens is it becomes a major burden that you carry for the rest of your life. You always think to yourself, "Oh, I wish we could have done this or I wish we could have gone on holiday here." Well, it is fine wishing. But if it is possible, I always suggest, "Do it." That is very,

very important....If it is possible and there is a way of doing it, then do it. Then you will not have that regret.

Marriages were severely strained because parents had no time for each other. They were so busy concentrating on the ill child that they neglected each other and their marriage. Parents had little, if any, time to be a couple. As one mother said:

The week that it is my turn to do nights, he tries to go to bed fairly early. Same when it is his week to do the nights, I try to go to bed. Sometimes it is like you don't see each other because you go to bed at 9:30 and that's it [laughs] and you are asleep.

Moreover, sometimes parents were even unable to sleep together, because one or other of them needed to attend to the child during the night. Some parents drifted apart and became more like companions or friends rather than lovers.

Even strong marriages were shaken by the effort and work required to stay intact under the difficult circumstances. One father noted:

Actually we had, have a very good marriage. But something like this will push it to the limit. It will just magnify all the negative things till they reach a point where it becomes a problem, something that must be addressed....Having a family like this just exacerbates everything, magnifies it and makes it all worse. Little cracks become truly huge gaps. Sometimes you just get tired of everyone.

All parents acknowledged difficulties in their marriages. Most parents tried to make the best of the situation and strove to keep their marriage and family intact. Parents had varying amounts of success in their struggle. Communication was the central key in the most successful marriages. These parents worked hard at developing strategies to improve communication within their marriage. Communication between parents was an important intervening condition that will be discussed later in this chapter.

When families remained together, parents usually shared responsibility for the necessary tasks that accompanied their child's illness. In the family where both parents stayed at home full-time, both looked after the child, although the mother always gave the child his bath. Where one parent worked outside the home, the parent at home usually had total responsibility until the

other parent came home and then the work was divided. Sometimes parents had preferences about what jobs they did, such as father bathing the child or mother giving vitamins through a tube, but mostly they just did whatever needed done. One family developed a schedule for nights, so that one parent could sleep:

Up until about 6 months ago we didn't have a schedule, so it seemed like we each went until we basically said, "That's it we can't take it anymore" and crashed. Whereas now with a schedule, with a program where we can actually follow day-by-day whose turn is it, there is a lot less frustration...at least one person gets a decent week and then the other person gets next week half decent.

Most families also shared responsibility for household chores. In some cases, traditional roles within the family changed, such as cooking dinner or doing the laundry:

Mother: And I always did the laundry and that is a BIG change because I don't even usually fold. I don't do anything as far as laundry goes. I've completely abdicated that and said, "It's yours."

Father: So, I fold laundry and put laundry away. I'm almost to a point, "Can you show me how to sew so I can do all the sewing for us?" Because if I've got a rip in my jeans or anything like that, it is sort of like "How do I fix these?"

Mother: Yeah, because if I don't do it right that immediate moment, it never gets done.

Father: It gets put on the back burner and sits down there until you go, "Well, I need a new pair," then so you go and buy a new pair.

Mother: Our roles have really, really changed.

Another change in roles involved a shift to more egalitarian decision-making between some parents. This was not always easy as one father explained:

I was carrying a massive load and I was able to share. It was just because it was something I was always used to doing. But with this crisis, it was getting bigger and growing bigger and with the panic attacks, I was finding that I was just not able to cope. Because they were a sign that there was something going wrong and I was able to then transfer some of it to Helen. But even then it took a while, because I had to make sure Helen was competent....It was hard to let go....If there are any major issues we try to talk about it. I try to get more input as well by bringing Helen more into the decision-making process. In the past, I would make those decisions.

Most parents shared responsibilities, but one family functioned differently than the others

- the parents clearly divided the responsibilities. In this family, the mother was wholly

responsible for the ill children and the father had little, if anything to do with them. There was a sibling in this family and the family was divided into two "mini-families." One consisted of the mother, the ill children plus the sibling; the other included the mother, the father, and the sibling. The father explained that his wife "provided the caring" and "worked towards doing the children," while he "worked towards just financing it and we've sort of parted. We both run two separate worlds." He managed his life by separating the personal from the financial aspects. He expected to deal with the "family issues" once the finances were stabilized:

Very honestly, I would say I've separated myself from my children and the issues of my children. I don't get involved. It's not - I think maybe I told you this before, because Sarah [older daughter], I adored Sarah, but... It's just - to make everything happen NOW that has to happen. I just can't be weighed down by that. So, I just choose to say, "Well, deal with it, Karen, because I have other things to look after."

In this family and in families where the parent caregiver was living alone, single caregivers expressed feelings of being more weighted down by the responsibility than if they had a partner to share it.

As previously discussed, a couple of families were considering placing their child in a care facility, such as a group home or long-term hospital, in order to keep the rest of the family intact. Not all families were able to remain together. In one family, the father left his wife and child because he was unable to support them financially. He believed that his family would be better off on welfare.

For some parents, the child was the compelling reason for staying together. They believed that an intact family unit offered the best opportunity for helping the child:

Mother: I think it is more important that parents should be together when a child is like this. They need more love, so they [parents] have to be together or they won't get peace.

Father: Or else we have to stick together for Mike. If one of us were to take off, the pressure on the other person would be just too great. They would not be able to handle it. It has got to be a team effort. There are no ifs or buts or anything about it. For us, Mike has actually brought us closer together.

The economic reality of the diminished income that would follow if a couple separated was also a deterrent in at least one case. The reason for another family to stay together was their house. The work the house required gave the husband a focus other than the child. In addition, the large size and scenic location of the house provided enjoyment for this family. Family members could have privacy when they needed it - unlike families who lived in smaller houses or apartments.

Although there were only a few siblings (four children in three families), siblings were strongly affected by the families' focus on the ill child. Everything in the sibling's life was dependent on the ill child. Families could not do anything on the spur of the moment, such as taking the sibling for a walk in the park when the child was at school. Even then, they had to plan for the possibility that the child would take ill and would need to come home. Participation in activities outside of the home, such as ballet or swimming classes, was difficult for siblings. One mother explained her dilemma:

I can't even send her [sibling] to pre-school because I cannot get Amanda ready for school and take her [sibling]. I couldn't, especially now. Amanda only goes in the afternoons. How would I get Janie to pre-school? I'd have to get Amanda ready. Heavens it would be a nightmare, and drive!

A couple of families enrolled the sibling in what, from an outsider's perspective, seemed an excessive number of activities. For example, in addition to school activities, one sibling attended softball, Brownies, and gymnastics every week. Another played soccer, took music lessons, attended Brownies, and so on. These parents were trying to involve the sibling in activities to compensate for the lack of a "normal" family life. However, the extra activities and the time they demanded added another dimension of stress in parents' busy lives.

Siblings had to follow routines that fitted with the ill child, rather than with their own wishes. For example, if the ill child went to bed at 8:00 o'clock, then siblings usually had to be in bed as close to that time as possible so parents could have a couple of quiet hours and could get

to sleep "at a decent hour too." While parents tried very hard to give the sibling "as normal a life as possible," the reality was that life could never be normal for the siblings while the child was still alive.

Parents tried to give siblings as much attention as possible, but their focus was on the ill child. Siblings did not get the amount of attention that the parents would have wished. Even when parents tried to limit everything else, such as turning off the telephone, and sat down to cuddle with the siblings or play with them, that time would be interrupted if the ill child needed attention. In such cases, siblings would get angry because they could not have their parents' undivided attention.

Parents also encouraged siblings to help a little with the child's care because they wanted to foster a positive relationship between the child and sibling. However, parents were clear that they, and not the sibling, were the caregivers:

The biggest challenge for her [sibling] is to make sure that she remains a kid....Amanda [ill child] is not her responsibility. She is to be a kid. We can look after Amanda and we look after her [sibling], not the other way around. She does not look after Amanda.

Parents often expected more from siblings than they would normally expect from a child of the same age, and some siblings seemed to accept the challenge:

We can talk to her quite well, but that's a lot, in part, because we expect her to listen as well. We don't have the opportunity to explain it 10 times over. She has to get it as quickly as possible because we don't have the same length of time to discuss an issue. It has to be handled, taken care of, and she has an understanding - all at the same time. Those sorts of things. Like other friends of ours - there are definite differences in what Janie is expected and knows what she needs to do.

The sibling's relationship with the ill child changed. Siblings lost a friend and a playmate as well as a sister. It was difficult for them to lose those relationships. Siblings were not able to play with the child as they had once done. In addition, younger siblings took on the role of "older sibling" because the child had regressed and was unable to function beyond a basic level. For example, a younger sibling would assist the child to draw a picture. Parents found that they

needed to work at fostering the child-sibling relationship. In some families, they received help from a child care worker who strove to include both the child and the sibling in activities.

In summary, *focusing on the child* meant that a family's time and efforts were redirected. Changes often occurred in traditional roles within the family. Family members increased their attention to the ill child and, consequently, reduced attention to other members of the family. Lack of attention to siblings and spouses created some difficulties for all families.

Putting Life on Hold

For many parents, their own lives went into a hiatus when the child became ill. Parents were in a state of limbo. They talked about feeling that they were "on hold." Plans seemed irrelevant because there was no future that included the child. Neither could they return to their previous lifestyles. As much as they wished the illness had never happened, parents could not change the fact that the child was dying. Parents put their own lives, and that of the family, on hold so they could focus on the ill child.

Many parents shelved their previous identities outside of the home because they remained at home with the child. Mothers perceived that they were expected to stay at home with the child, yet not every mother was comfortable in this role. The expectations of others added another stress to those mothers who felt they could not provide constant care to their child. In one family, the mother chose to return to work after spending three years at home with her child. She hated being confined to the house and was very depressed. She felt "like a prisoner in my own home." Another mother upgraded her education so she could work outside the home. Both women gained a new lease on life when they re-entered the work force. They dreaded any future event that might compel them to stay at home full-time.

In some families, parents continued working in the early stages of the child's illness. As the child's condition worsened, however, at least one parent gave up work. The parent who stayed home with the child was normally the mother, but in three families, the father could no

longer work because of the effect his child's illness had on him. In a few families, parents were self-employed and chose to give up their business. These parents stopped working or took a position elsewhere as an employee. In several families, parents went on welfare as their only means of support. Giving up work was not an easy decision for most parents as this mother and father acknowledged:

As she progressed or as she had more seizures and more problems, it got more and more difficult to do it. I had to finally say, "That's it! I can't, I cannot do this anymore!" It was a difficult thing because I really, really enjoyed that work and I had been offered several positions with companies that put on conventions and things and I wasn't able to take them. I would have loved, loved to have done it, absolutely loved it, but there was just no way. So, that part went by the wayside.

Yes, a major change for both of us, because we both had been working for a long time and very, very hard at it. So it is difficult to sort of put the brakes on. It was a question of something has got to give, because we had this crisis, and it was affecting everybody - it was affecting Helen, it was affecting me and affecting our son. You can't have the work pressure with the type of work I had; very, very high pressure all the time, so something had to give. So, I decided my family was just too important, and I walked away from work.

Some employers were very understanding and facilitated continued employment, or negotiated long-term disability, so that parents could spend as much time as possible with their child. Flexibility in the workplace was extremely important for those parents who continued to work. One father said:

I have the flexibility with my job that I can pretty well, if I have to, get home [early] which is really, really important for our situation. I sometimes wonder what we would do if I didn't have such a good and flexible job. I don't know - if I had a job that relied on me selling something or me keeping on top of financial markets, or who knows what, but something that I really had to be on top of all the time and the amount of my compensation depended on how much effort and time I put in, then we'd really have a problem....For certain, both of our lives would be a lot more stressful. So the fact that I have the job I do really makes things a lot easier.

Other employers were not as flexible and parents were either asked to leave or they made their own decision to leave.

Families placed their financial plans on hold because of the situation. In many cases, families lost not only their accustomed income, but also potential income. Some parents had

expected the mother to return to full-time employment once the child reached school age. That was now impossible because the child was ill. A few parents could not work overtime or had to change shifts so they were home in the evening, thus losing the pay differential. All families were less financially secure than they had expected to be at this stage in their lives. For example, parents in one family had anticipated reducing their mortgage and moving to a bigger house. But their goals were based on having two incomes. They were forced to sell their house and move into a smaller one. These parents, like others, felt they would never achieve their life goals and dreams. As one father said:

You base your lifestyle on your combined income and then it's gone. But the bills keep coming in because you bought things, such as a house and car, with the expectation that you would have the income to pay for them.

Many other things were neglected or put on hold as parents faced a relentless situation. They were always "on call" and were frequently exhausted. Often, the child did not sleep at night so the family could not sleep either. Or the child screamed incessantly for hours despite all efforts to soothe him/her. Parents gave up their previous pursuits. For example, one father used to do the mechanical work on his own vehicles, but now he used garage services as a convenience. Recreational activities were often abandoned. Parents had little time for relaxation because they devoted their time to the child's care. Even reading a book became almost impossible because of lack of time, and also because parents were unable to concentrate on anything but their child. In addition, parents had to think carefully about the potential consequences of their attempts at relaxation. One father said:

I used to enjoy a rum and coke or a nice drink in the late evening, where we would sit down and have a bottle of wine or something. We can't do that, because if it is your turn [to get up with the child]....You don't want to be in a state where you are not fully aware. Or conversely, you don't want to have one too many drinks so you can't drive. So I mean you can't even have two beers in case you have to go. That is one thing that really has been - there are times when you would like to just sit down and have a nice bottle of wine and not have to worry about leaving the house or doing anything and just enjoying and relaxing the whole evening with it....So those are just little perks that you think twice about doing. Before, you wouldn't even have thought about it. If someone brought some

beer over you would be drinking. Come home from work on a hot summer evening you have a drink. But now you don't, you just don't.

Parents were also restricted in their sports activities. Most parents did not have the time for practices and games on a regular basis. They were needed at home. They perceived that they could not take part in sports where they might get injured, such as hockey or skiing. As one family explained:

Mother: We used to be very active. We used to ski.

Father: Yeah, you don't do any sports that could injure yourself. Because, like friends just phoned up, "Do you want to go skiing with us for the day, come with us?" And I go, "No." I don't ski because I don't want to injure myself. If I injure myself I can't help. So you just don't do things that could injure yourself. You stick to your sports or things that are as safe as they can be.... You do things a lot differently, because when you are injured you realize how little you can actually do. And then when you start to feel better, how much you have to pick up because the other person is so exhausted [because they had no help in caring for the child].

If there was any free time, the type of activities families could participate in were restricted. They stayed at home a lot more than they used to before the illness. Simple outings became difficult. They could not go out to restaurants, the park, or the playground because of the child's symptoms or behaviours. Watching a movie caused seizures in some children. Or loud noises bothered them and they "ended up screaming, with uncontrollable screams." The more difficult it was to "pack up and go," the more reluctant families were to make the effort to go out. Most families stopped taking the child out, or at least reduced the frequency of outings. Instead of an outing as a family, one parent stayed home with the child while the other went out. Siblings became used to going places with only one parent. However, when parents went out with only the sibling, they were sensitive to other people's perceptions that the family only had one child. Some parents felt guilty, as if they were disowning the ill child.

In conclusion, families were in a state of limbo. They could not plan for the future because the future did not include the child. Financial plans were put on hold. For many individual family members, as well as for the family unit, previous activities were no longer

possible. They were relegated to the background while care of the child took precedence. Family members shelved their own needs as they struggled to manage the work that accompanied the child's illness.

Getting Rid of Excess Baggage

Life and relationships outside the immediate family continued. However, most parents discovered that they could not maintain their previous relationships. While extended families and friends often withdrew from families over time, families themselves also disengaged from relationships with others. They focused so much time and energy on their child that they had nothing left to give to others. As time went on or the child got sicker, families needed to "take stock" and decide where they would expend their limited energy. For example, in one family the maternal grandparents were heavily dependent on emotional support from their daughter. The daughter struggled to support her parents while caring for her ill child and immediate family. Eventually, she broke off contact with her parents so that she could focus on her child. This mother said:

The sicker she [child] got, you have to make decisions. Something has to go. You can't keep on doing the same things all the time so you have to decide what is really important and then you do...you just have to get rid of that stuff.

And a father in another family was quite clear on what he could and could not do:

I found out now, in the last year or two, that I have to start drawing lines with people, and say, no matter who it is, even if it is my family, no matter who, "This is the maximum I can afford to do for you. After that, it is up to you. But this is the maximum." Sometimes, it might be only for me to move an inch, sometimes, it might be a foot, but that is the maximum I can do. I will lay it on the line and say, "This is the maximum I can do." Because with us we have to. Mike takes up a lot of our energy, thoughts, everything, and we do not want to be burdened with everybody else's problems. That is why I said the road becomes lonely as well, because you find everyday your reserves - especially when the stress really gets on, when Mike gets really, really ill - you start going into your reserves. Then you just don't want to hear about anybody else's problems. You don't have the energy to deal with it....It is always a question of stress, because there is enough, our plate is full, we do not need anything more. We decide ourselves because we know exactly what we are faced with. We know how much we can take and there are days when we say it is not worth it.

Parents also withdrew from other families as the child became more ill or needed more equipment because it was difficult to take the child anywhere. One father explained:

Now it means the whole van is full just to go out for the day. So, to visit family and friends is hard because it means you have to load up the van before you go out and then bring her home and empty it out as well. It is not bad for day trips, but if it was for a weekend or anything of that nature, we almost have to ship everything up ahead of time.

Maintaining friendships with other families who did not have an ill child was difficult for most families. Friendships required a commitment that families could not make because of their focus on the child. A father said:

Yeah, relationships with them change, partly because we are unable to fulfil the other end of the friendship as well. I mean if they phone up and say, "Do you guys want to come over?" we basically have to say, "No." So it is hard to maintain that relationship when you are unable to go down the other end of the street basically.

Maintaining friendships was also difficult because families no longer had things in common with their friends, as this mother explained:

So you lose touch with what other people are doing or...what being a family with healthy children means....Sometimes you get so sick and tired of - like this is what your life revolves around. You find it very difficult to talk to your normal friends. We have to work very hard at staying current and up to date and trying to think of interesting things to talk with our friends about because we can't say, "Amanda went to ballet school."

In fact, some parents did not even know what kinds of things families with healthy children did.

When their child was booked for respite care and the family could have a weekend or a vacation without the child, these families needed to learn what was available to them. They did not know what they could do or where they could go, because they were "not practiced at it."

Another reason the families tended to stay away from families with healthy children was that the ill children could not interact with other children:

With the fact that we have two disabled children...we don't get together as much with other people that have children because there isn't that natural interaction between them....So that's a bit difficult to deal with at times, because it's sort of sad that - you can't blame your kids...they're different, they don't interact and you can't force them to.

In addition, it was too painful for some parents to see other healthy children when their own child was ill. One mother explained:

There was a small period of time where I didn't want to be with our friends. It wasn't just them that pulled away, it really was us too. I couldn't see other children her age for a couple of months. It drove me crazy because I would look at them and their parents would say things like, "Oh I wish we had never had kids," and I would get so mad because I'd think, "You should have had a child with this disease, you don't care about them. Why did we have a child with this disease when we love her so much?" But seeing these other little five year olds running around when she started school was so difficult because the gap had suddenly widened. She reached her peak at three and so from three to five it was okay. But once you start going over that five mark, the gap just got wider and wider and it was so painful to see other children, her friends, who were progressing and so I didn't - there were children around, but I didn't want to get to know them.

The decision to disengage from others was usually a difficult one. Parents often struggled for months or years to "do everything." They were torn between their child and extended family or friends and they anguished over what they should do. Yet, withdrawing allowed them to focus on the child. Those parents who were able to get rid of the "excess baggage" found some peace and relief in their lives. But sometimes they also became more isolated and lonely.

Picking up the Badge of Disability

Some parents accepted their child's disability and prognosis better than others. All parents acknowledged that their child was disabled. However, each deterioration in the child's condition caused some parents to redefine their understanding of disabled. One mother explained how her understanding changed:

I'm just now coming to terms with the fact that I can't take Sarah [out] anymore. That's been the next step for me. You accept every level and it gets harder and harder. You take more and more equipment with you. You are more prepared. To get to a point where I have to say, "I can't take Sarah anymore, she's better off staying at home," to me is a big admission of her weakness now.

Parental recognition that the child was disabled was important. But redefining the child as disabled was critical. Families were thrown into a new world of disability that they knew little about. They did not want to be part of this world, especially at first. Some parents struggled to understand this new world. They felt a need to reflect on their own lives and beliefs about what

being disabled meant. They reached an understanding that their child was disabled and accepted the limits that accompanied disability. While they realized that the label "disabled" held negative connotations for many people, they also viewed the label as a possible advantage. For example, they wanted their stories to be heard, but they did not have the time to tell everyone. When others saw the child in a wheelchair, parents did not need to say much more. Parents also used wheelchair-bound celebrities as examples of the positives that could come with disability. For these parents, *picking up the badge of disability* was a "matter of survival." It enabled them to accept the child's condition. It also opened doors to services and organizations that provided assistance. These parents then found it easier to manage their lives.

Some parents, however, did not accept the extent of the child's illness. They "stayed on the fence." One father acknowledged that his son was disabled and terminally ill. Yet, when the child was expected to die, the father left. The child's grandmother and aunt stepped in to be with the child. The father could not think about a funeral and refused to deal with arrangements. The child unexpectedly survived this critical episode and returned home. But the father still would not talk about the possibility of death.

In two of the families, a child in the extended family was also afflicted with the illness. According to research participants, the parents of these other children did not accept that the child was ill. They were "in denial." These parents did not accept the help that was available for their child. They continued to treat the child as "normal" despite evidence to the contrary. One child had frequent seizures, but his family refused further testing to ensure a definitive diagnosis because, "He is not like his cousin. Yes, he might have seizures, but he is very different." Research participants perceived that the children were suffering because the illness and its implications were ignored. For example, one child was "forced to go to a regular school" where he was teased mercilessly by other children who did not understand that he had a disability.

For most families, events settled down after the hectic, initial few months of the illness. They returned to a "semi-normal" life. Most parents quickly recognized, however, that their lives "could not be normal" because their child had an NLTI. Normalizing meant keeping things the way they used to be or the way other people did it. This was not possible. For example, one family had a motor home. They tried one weekend to "take our world with us." Their child "almost caught pneumonia" and as the father said, "It's just not worth it. That was sort of our stab at, 'Okay, we'll just take our world with us and we'll go when we want.' But we can't."

The situational context prevented most families from appearing like other families: the child's disability was often visible, families focused heavily on the child, and they put their own lives on hold. In most cases, the more complex the care that the child required and the more equipment needed, then the more likely families were to acknowledge an abnormal lifestyle.

Most families attempted to make their lives as normal as possible within the constraints they were under. Parents strove to keep their lives "low key" and as "less stressful as possible." Some families used respite care for the child to ensure that they could spend some "normal family" time with siblings. They recognized that life was not normal when the child was around.

Parents who did not redefine their child as disabled struggled with creating meaning in the child's illness. They were unable to accept the situation and had difficulty managing the emotional work involved. These parents wanted a "normal life" and were less able to let go of the past and live for the present. Other parents who picked up the badge of disability were more comfortable with the situation. They just "did their best" and found it easier to create positive meaning in the situation.

Creating Meaning

The third dimension in *navigating uncharted territory* also occurred throughout the process. Families continually created meaning as they learned of the child's diagnosis, faced frequent crises, and entered unfamiliar territory. Parents struggled to understand why their child

was dying. They also needed to find ways to make their child's life and inevitable death worthwhile. Parents desperately searched for ways to give the experience some meaning. Creating meaning was a highly significant aspect of the families' experiences. Some parents were more successful than others at creating meaning. Those parents were able to recognize their multiple losses, but still make the most of what they had. They often took active measures to improve the quality of their lives, such as disengaging themselves from relationships that were harmful to the family in some way, or spending quality time with their child and any siblings. In many cases, these parents perceived that their family quality of life was good because they enjoyed life more than other people.

Other parents felt threatened and challenged by the child's illness. They could see little good in the situation. The threatened parents were prone to depression and anxiety. They perceived that the quality of their lives had diminished. They expressed the view that they were only "existing, not living." These parents were sometimes angry and bitter, and felt that life had given them a "raw deal" and that life was unfair. These parents were more likely to find fault with the health care system and were more likely to blame God or other people for their troubles. They were also less likely to feel that they had grown in any way from the experience. They often had a hard time visualizing any future and did not have the ability to see any good in the experience. In situations that were perceived as more threatening, marital relationships were strained or finances were limited, and the particular parent was having a difficult time dealing with the situation.

Parents were searching for ways to comprehend the situation. They created meaning in the situation because it enabled them to accept the child's illness. Only then could they find the strength to continue living with the knowledge that their child was dying. Parents created meaning in a number of ways: *taking one day at a time, reframing the experience, reaffirming*

faith, and *participating in research*. Each of these strategies enabled parents to find meaning that was acceptable to them.

Taking One Day at a Time

Parents thought about the child's illness every day. The threat of death never went away, but most parents stopped dwelling on it a few months after diagnosis. All parents took one day at a time. Thinking only about the present and the near future facilitated hope and limited the uncertainty associated with the situation. Parents tried not to think about many of the problems that might arise in the future. Looking far ahead overwhelmed them because the problems seemed "so big." Parents found it easier to "wait until you get there." They dealt with each day as it came and faced most difficulties as they appeared. As this father explained:

If we can make it through to the end of the day, that is all that is important. Worry about tomorrow when tomorrow comes. If we go one step at a time and one day at a time, then we will be able to accomplish one year at a time.

Paradoxically, some parents who "dealt with each day as it comes" also planned ahead to enhance their management of resources. For example, parents did not know how long the child would survive, so some "went to the extreme" and planned accordingly. One family discussed "the worst case scenario, that we have to survive the next 30 years." These parents worked out the amount of money they would need over an extended period and organized their lifestyles to make it work financially. They cut back on many activities, such as planning to take "mini vacations instead of major vacations," to conserve their limited funds.

Parents found that thinking about and planning for certain potential difficulties allowed them a degree of respite when inevitable problems arose. Forward thinking also allowed them to put resources in place before the child was desperate for a piece of equipment. One father explained:

There are certain things you've got to start thinking about. We've been talking about the porch lift that we've got to get for Mike. We know there are funding agencies - there is other help out there. It is a matter of getting things going. Also, some of these places only

meet once in three months or once in six months. So you've got to start working ahead as well.

Some parents appreciated that their child was "only around for so long, so we make the best out of every day." They learned to "enjoy the day." Simpler things, such as a smile from their child, gave them pleasure. These parents took delight in focusing on their child's abilities and working with those. One father explained:

You would take a lot of those [normal activities] for granted. But now the focus is on quality of life. It is a totally different focus. Whereas in the past it was maybe education, maybe trying to provide everything that you can. But it would be different then because for all purposes your son could do a lot more different things as well. Now you are restricted. Now you've got to scratch your head and say, "He is not normal. What can he do?" and focus on those things and not say, "Well, he can't do this." That's fine, we know what he can't do, but let's find and focus on things that he can do. He is blind, he cannot hear, he cannot walk, he cannot talk, which is fine. But what can he do? [If] he can hear you, [if] he can sense your touch. Let's work with those two major senses and take him out as much as we can, make sure he can listen to all the birds and the bees, make sure he can feel the sun on himself, make sure he gets to hear all the other children laughing, make sure we take him out to a water park nearby as often as we can so he can hear all the screaming and the laughter and everything else.

Taking one day at a time helped many parents "live with what you've got." Living one day at a time, however, was also very difficult because it was a constant reminder that the child did not have a future. This mother said:

I live day by day and it's a terrible, terrible, terrible way to live. I would never ask anybody to live day to day. You have other parents that can think of high school and college and grandchildren. I don't even think of that.

Certain days, such as the child's birthday or Mother's Day, were forceful reminders of future losses and were especially difficult for every family. At the same time, these special days were sometimes viewed as "milestones" and indicators that the family had survived another year. "That in itself is a sense of achievement. So it is almost like we pat ourselves on the back again and we just keep moving forward."

Taking one day at a time helped parents limit their uncertainty and distress. They were able to focus on the child and the current situation, rather than worry about the future. They

could also appreciate each day and could enjoy the simpler things in life. Living from day to day was also difficult because it reminded parents that their future could not include their child.

Reframing the Experience

Some parents looked for the positive side in their experiences. They explained that it was vital to look for positives because "you can't survive otherwise." The positives were not always easy to find. Parents often had to "really look for the good things," such as the "wonderful people" they met because of the child's illness. They also surrounded themselves with others, such as home care nurses, who were positive. "To be able to survive this, we need people that are very positive, that are going to help us." These parents did not want sad people around them either. They wanted laughter in their lives and they looked for humour in their situation. Parents also appreciated a good sense of humour in others. Professionals who were not afraid to laugh and joke with families were perceived as "really great" and were welcomed by most parents.

Parents who looked for the positives did not feel as threatened by the situation. They perceived the situation as an opportunity rather than an insurmountable challenge. These parents tended to see their child's illness as a potential growth experience for themselves. They gained more confidence in themselves and found inner reserves of strength that they had not known they possessed. As one mother said:

I'm happy with where we are at now. Yes, there is still stress with Amanda. But, it has been a real growing experience. It has been lots of terrible things - coping with all the seizures and the fear and everything - but once you get past that and you start to see the good that comes out of it, or maybe if you like yourself at the end of it, somewhere along the way it doesn't seem quite so horrible.

For most parents, positive reframing also involved making downward comparisons. No matter how difficult their own situation seemed to an outsider, these parents could always find someone else who, from the parents' perspective, was worse off. For example, one mother compared her son with other children in hospital:

When we used to go to the hospital, I used to see the other children and how they are suffering - he [child] can't talk or can't do anything, but otherwise he is okay....He is still here, and I see him 24 hours.

Families with one ill child reacted with consternation at the thought of having two ill children.

But families who did have two ill children found other situations that seemed more difficult than their own.

Parents had a strong desire to find some meaning in their child's life. Some parents perceived that the child had a positive impact on the child's peers at school. For example, in one school, the child's class was perceived by teachers as "one of the most exceptional." The child's presence had taught classmates valuable lessons about caring for others and about differences in people. These students became more caring and empathetic. They also changed their views on how to deal with others with disabilities. Even teenagers in the school who were "more wayward" were more sympathetic to one child. They changed their behaviour in her presence, such as turning down loud music or extinguishing cigarettes. The parents of other students commented to school staff that the experience of being in class with an ill child was beneficial to the students. They were hopeful that the students would continue and extend these changes throughout their lives. For the child's parents, making a difference in other people's lives gave meaning to their child's life.

In many families, there was a change in values or in how the family enacted their values. Often, their focus changed from material things to focusing on quality of life. They lived a "simpler, less complicated life." They found that their values changed or were reaffirmed. For these families, their previously "normal" lives were not necessarily what they wanted any more. Parents slowed down and appreciated the "smaller, finer things in life." They made changes in their lives to accommodate their values. They examined different aspects of their lifestyles such as their diet. For example, one family gained knowledge about the effects of diet and the environment on health and started to eat fewer meat and more vegetarian dishes.

Some parents also became more empathetic to others and were the ones who always tried to understand another person's point of view or reaction. One mother said:

I might be a different person if I hadn't had these kind of children....I think it's made me grow up a little bit better - to gain a little more perceptive of what people are really like and what life can be really like.

These parents endeavoured to understand why extended family and friends withdrew from them, or why the physician suggested that they should just let their child die. Instead of becoming angry and bitter, they reframed the experience in a positive light. They often justified the actions of others and found reasons for their behaviours. They also used humour in their interactions with others and were more likely to look for the best in their situation and not dwell on the difficult aspects.

Parents who reframed their situation in a positive light were more satisfied with the experience. They found meaning in the child's illness that allowed them to continue caregiving. Moreover, some of these parents viewed the experience as an opportunity for personal growth.

Reaffirming Faith

Parents struggled with many existential questions as they tried to accept and create meaning from the child's illness. Many experienced deep spiritual pain. They searched their souls for answers to why the situation had happened to their family. They raised spiritual questions about who or what was to blame. Parents railed against the perceived unfairness of their situation. They questioned if the child had been given to the family by a "higher body" as part of a larger cosmic plan. Parents perceived that health care providers were often uncomfortable with this spiritual and moral struggle. In some cases, professionals told parents that their thinking was wrong or misguided. Yet, parents emphasized the "normality" of asking oneself questions such as, "What have I done to deserve this?" Moreover, they asserted that this struggle was an essential component of finding meaning and achieving acceptance.

Most parents acknowledged some religious affiliation and stated that religion or spirituality had a place in their lives, at least before the child's illness. Their faith helped some parents manage the situation. One mother said:

I do believe in God and I think I always have believed in God....I said, "I have too much guilt. I'm not to blame for what my son has....So, I just asked God to take all this guilt that I had so that I could continue on. To give me the strength to do whatever I have to do with Ian. The next morning I woke up and it was like - honestly it's weird, it's like this big weight had been lifted off of me. It was weird. I even had to phone my pastor and say, "This is too bizarre. I can't handle this because I feel too good. I shouldn't feel this good. I just separated from my husband. I have a terminally ill child. I'm in a new apartment, I just shouldn't feel this good."

Faith also helped parents find peace in themselves and with God. A few parents perceived that they were "serving God everyday" by caring for the child.

Sometimes parents lost their faith temporarily, but eventually reaffirmed their faith. They may have questioned why this was happening, especially in the beginning, but they did not blame God for the child's illness. In some cases, parents thought that they had been "chosen" to have a child such as this and they saw the child as a "gift" from God. These parents prayed to God regularly. However, they did not always attend religious services or receive support from their spiritual community. Families who wanted to continue their participation in religious ceremonies, such as mass or evening prayers, often found it very difficult to attend such events. The effort involved in preparing to go out with the ill child, or the difficulty in finding child care often made participation impossible. As one mother said, "It takes two hours to get ready to go out. It's just not worth it." Families sometimes lost some of their support system because they were unable to participate in their church.

Parents who reaffirmed their faith in God accepted the situation and were more satisfied with their lives than parents who had lost their faith. A few parents gave up on God. They were angry with Him. They felt "discouraged" and "let down" by their faith and church. These parents blamed God for what had happened to the child. They were very bitter. Often, they felt

personally punished. Yet, they could not understand why they were being punished. Moreover, they believed that their sick children were being punished, yet the children definitely did not deserve what had happened to them. According to these parents, the whole family was being punished and was suffering. They could not see any reason why God would do this to anyone, let alone to children. They had lost their faith and could find little meaning in their experience.

Participating in Research

Parents were adamant that their children should not be used as "guinea pigs." Yet, they agreed to participate in some types of research. Sometimes they wanted to raise awareness about the child's illness or their experiences. "I think with your study [current study], we want to help so that other people know." Not only did they want individuals to understand, but they wanted broader system understanding, such as the health care system and society in general. They also hoped to change societal views about disability, the needs of disabled people, and the contributions that disabled people can make to society.

Some parents consented to experimental treatments for their children or to their participation in basic research. They wanted to help scientists find a cure for the disease or at least make it better for other children. One father explained:

That's one reason I'm glad Ben's here. He's able to help. They can take a skin sample and they can check the DNA and work on and grow it and go on until they find it. I'm glad he's able to help that way.

Even when participation in research was difficult for parents, they were so committed to helping others that they were willing to suffer themselves. During a second interview, one father said:

Last time you [interviewer] left, I had gut pains for a week. It [interview] brought back every memory. And just sitting here this morning. I WANT to co-operate because inside of us - we want to help other people that are hurting like us. That's [the] bottom line, why we're participating. So if I have to go through this little bit of anxiety and pain, then that's what I have to do. But that's what this does to me. Right to my guts.

Parents used participation in research as a way of helping families in similar circumstances. They also hoped to find a cure for the child's illness, even if it was too late for their own child. Finally, participation in research allowed parents to create meaning from their child's illness.

Holding the Fort

Holding the fort was the fourth dimension in the process of *navigating uncharted territory*. This dimension did not start until families reached some stability. The first few months, even years, following the child's diagnosis were tumultuous. Eventually, there came periods of time when the illness stabilized and families reached plateaus of relative calmness. For many families, these were times when they felt more in control of the situation. They focused on maintaining stability and preventing further deterioration in the child's condition. Treatments or caregiving became routine and just a part of life. Parents gained increased confidence over time and were able to juggle multiple tasks. However, families still often found it difficult to fit everything in, even after years of practice. As the child became sicker and required more complex care, performing multiple tasks became increasingly challenging. Parents worked very hard at *holding the fort*, but they often faced crises that disrupted their stability.

During the periods of relative stability, families continued to live with uncertainty. They were living on the edge, waiting for the next crisis. Their situation was still unrelenting. They were exhausted and lived a "treadmill existence." Life as they had known it remained on hold. Caring for their child was a 24-hour-a-day, 7-days-a-week responsibility for families. They felt "on call" every minute of every day, usually even when someone else was caring for the child. They were always listening, watching, waiting in case the child choked or vomited, or the other parent or a professional needed assistance. They were always anticipating that they might need to rush the child to hospital. Even a quick two-minute shower was often impossible. Parents were never able to relax. They had to rent a movie instead of watching it on television. This way, they

could pause the movie; otherwise, they were continually interrupted and missed the program.

Even when the child was out of the home at respite, they were so used to being "on call" that they could not always relax. One mother explained it this way:

The odd time I don't even sleep even if she's gone now. Sometimes I don't sleep because I'm so used to waking up that sometimes I just end up waking up anyways. And I don't even have to, but I do. That's why she [psychologist] says you're in such a cycle with it that you're just - it's like an automatic thing. It's like firemen. You know, firemen go to work. They've got those four days they're in the fire-hall and they get woken up sporadically for fires and stuff. Well, that's like what we do with Alexa, but we don't get that break. They [firemen] get that break. They get to go home for three nights and sleep and then go for another four days and sleep or go three days, four days off. But we go, what is it...almost like 14 days straight and then we get a break, then it goes another 14 or 12 days then you get a break, so we're like right in there.

Parents lived by the clock as they tried to manage the work they needed to do. They co-ordinated and learned to live with the multitude of services and personnel that were required for their child's care. Parents provided most of the child's physical care, and they worked incessantly to promote the child's health. Many families also formed relationships with other families of disabled children because they gained support from such families.

Despite the rigours of managing their lives, many parents were extremely creative and innovative in finding solutions to the problems they faced. As one father noted with a smile, "Necessity is the mother of invention." Families were in uncharted territory. Often, neither parents nor professionals were aware of existing solutions because little had been documented. So, when parents came across a situation that required a new solution, they often developed "mind-boggling ideas" that solved their problems. Some of these ideas will be discussed under the strategy of *providing physical care*.

Families held the fort by using five major strategies: *living by the clock*, *co-ordinating services*, *providing physical care*, *promoting child's health*, and *forming new relationships*. Each of these strategies will be discussed in the following section.

Living by the Clock

Living by the clock was an important strategy that parents used to *hold the fort*. Parents viewed time as a precious commodity. They were very time conscious because "you end up with worse problems than when you don't have the time correct." They planned meticulously to ensure that the necessary work was accomplished. One mother explained:

So we can plan outings, but it means making sure we come back. We try and use the portable pump [for feeds], but there are days and times when it is not practical. It is easier rather to schedule short outings and come back. Some days when you would really like to go up to Grouse Mountain - but you know that from here to Grouse Mountain is a 45 minute drive and another 45 minutes for the way back. You've got to at least allow an hour or maybe an hour and a half while you are there, and with Mike it is probably closer to two hours. So we are looking at two to three and a half hours, maybe four hours. It means a lot of planning. Sometimes it means getting up at 3 o'clock in the morning to make sure he has enough nutrition prior to going...so you know you've met the target for the day.

Parents were frequently "clock watchers" even when they were out without the child. They needed to be back home at a specific time to relieve the child caregiver. Being so time conscious spoiled the enjoyment of having a break from caregiving:

You've got to get back at - the time is set for you. So in other words, you are leaving your company not when you want to, but when you have to. You sit there and you start looking at your watch. "Oh, we've got to leave in 10 minutes, we've got to leave in 15 minutes." Whereas before we'd say, "Okay, good night" and away you go. Now you've got to think about time as a factor.

Families learned the value of routines and schedules. Everything revolved around the child's needs and schedules, and families' lifestyles became very regimented. Some of this regimentation was externally imposed, such as their need to focus on timing of medications and feeds. They also used time regimentation as an internally imposed mechanism to help them manage and co-ordinate their lives. In addition, the children did not tolerate change well and they needed routines in their lives. One mother explained:

Days tend to be actually very routine, because that's the only way the boys have been able to tolerate it....They have to know what's coming and if they don't know, they get very - they can be very difficult. Time changes of the meal time - they don't tolerate that....They eat breakfast every morning at 7.30. Within 10 or 15 minutes or so around

that time, I have to have it ready. They get this little clockwork set up in their little bodies. It's good because then you know what to do with them at what time and what they might be needing at a certain time.

Time took on a different meaning for families. Parents became much more conscious of time. They were constrained by the schedules imposed from external sources. Yet, routines and schedules were also valuable in facilitating some control over the situation.

Co-ordinating Services

Parents spent a lot of time and energy determining what services were available to meet their needs. They often struggled to obtain initial services and then continued fighting to maintain or increase services as the child's condition deteriorated. Once services were in place, parents, usually the mother, invested many hours in co-ordinating services. Caring for the child was a full-time job for families. Yet, it also required a lot of effort to schedule appointments and personnel. One mother said:

Looking after Sarah and Nancy is a full-time job. Plus all the scheduling, plus all the keeping the staff happy. Planning. It's phenomenal. It's like running a small business. There's about 15 of them I've got working here.

Nevertheless, managing the child's care was very helpful for some parents. It gave them a sense of control, at least over some situations, when they had no control over what was happening with their child.

Families often had little free time. Their calendars were full with appointments. They received visits from many different health professionals, such as physiotherapists, occupational therapists, and nutritionists. Or they took the child to multiple appointments at clinics and with therapists. Scheduling and co-ordinating the many appointments was difficult and required parents to spend a lot of time making telephone calls. "The phone is usually ringing off the hook from 9 o'clock - therapist appointments, you name it, it starts at 9:00." Some parents were frustrated at the time wasted as they tried to co-ordinate services. They also noted that, despite

their best efforts, everything could be "wrecked by an unexpected change," such as a nurse calling in sick.

Some services were in the community and others were at home. Most families got some short-term respite. For example, in some families, nurses or child care workers came into the home; in other cases the child went to a respite family. In still others, respite care was provided in an institutional facility. Some families also received respite from a pediatric hospice. Parents struggled to combine these services in ways that were most beneficial to the family. Families were grateful for the respite and credited respite with keeping the family intact. One mother said:

But I don't want people to take away the supports that we have. If they do that, then we'll probably fall apart. Because, what we get now has made us [what we are] today. If they were to take it away from us now, then they'll just be making our family fall apart again. It has taken us a long time just to get to where we're at now, and we're finally comfortable with what we've got. So, I think as long as they don't take it away and keep it the way it is, I think our life will be okay because we have that help and that support. But if they were to take it away I think we would fall apart, because it's just too hard to manage.

Many families were dependent on professionals to provide some home care to the child. Parents attempted to schedule personnel to suit the family needs. In many cases, however, parents had little control over the hours that a health care professional was allowed to work. The government programs that supplied the workers often designated the number of hours and the shifts that families could obtain services. Families planned their lives around those of professionals. For example, an evening out was only possible if a caregiver was available. Parents were also at the mercy of professionals' schedules at holiday or vacation time. Sometimes parents were deprived of sleep because the nurse was sick or bad weather prevented her from reaching the family's home.

Having other people in their home required considerable adjusting for most families. For example, it was difficult for parents to sit down and have a private conversation or a cuddle

without planning it. They always had to check to see who else was around and when they would be leaving. Families lost privacy:

Yeah, they know if you are not feeling well, if you've got to get up and get a painkiller. We keep our painkillers in the cupboard with all of our daughter's medications....So if you have a back ache or a headache or whatever, you have to traipse out here, barely able to see, open up the cupboard, fumble around. And she is sitting there. It's like "Hi." Get yourself a drink of water, and she has learned not to - she used to kind of talk to me a little bit. I just - I'm half asleep, I actually walk with my eyes closed I've discovered. But it is a very different kind of life-style [having someone in your home].

Parents were creative in finding some privacy. One couple talked on the telephone during the day, so that the person in the home would only hear one side of the conversation.

Though families often relied heavily on workers in their home, not all workers were helpful. Parents needed workers they could trust and who could work independently. When parents had to expend time and energy ensuring that workers were doing decent work, the workers were often more trouble than they were worth. Families found it easier to accept workers into the home when they trusted the person, especially night nurses so families could get needed rest. Over time, many families became close to the professionals and were comfortable with them. When families lacked confidence in workers, sometimes they called a supervisor and asked that a worker be removed from the home. In most cases, their request was honoured. When parents found someone they could trust implicitly, they tried very hard to keep that person working with them. In turn, workers were usually very accommodating. For example, in one family, the nurse arranged her own vacation schedule to coincide with hospice respite care for the child so that an unfamiliar nurse was not needed to cover for vacation time.

When workers were present only for limited time periods, families found it easier to deal with having extra people in their homes. However, the sicker the child became, the longer workers tended to be in the home, and the more difficult it was for families to get on with their lives. A gradual increase in hours allowed families to become accustomed to having more people around. As long as workers did their job and did not interfere with the family's life, then families

more easily accepted them in the home. Parents occasionally imposed limits to ensure some quiet time:

He [husband] says he wants everybody out of here by 8 o'clock at night, or 9 o'clock at night, and he doesn't want anybody on weekends. That's fine. I rearranged the schedule so that I can try and work it so that there's no one around after 8 o'clock during the week or on a Friday. Or there's very few people around on the weekends. But as I said to him, "That's fine for all these people to go, but it doesn't mean that our children don't need any care after that time. So, if they've gone, then don't expect me to be able to sit down next to you on the couch because I'm going to be in there having to care for them."

Some parents found it easier to "do it myself" because of the immense work when others were involved and to ensure they maintained some privacy.

Families depended to varying extents on a number of services. Co-ordinating these services, while ensuring that family life continued as normally as possible, was a major undertaking. Parents devoted many hours to finding available services. Then, parents continually worked hard to schedule appointments, therapists, and caregivers. At the same time, they needed to accommodate to an increased number of people in their home. Parents needed well developed managerial skills to ensure everything was done. Professionals seldom appreciated the amount of co-ordinating work done by parents.

Providing Physical Care

While every family received some respite, the families themselves provided the majority of the child's care in the home. "Nursing staff come in and give us a break one and a half days per week, the other five and a half days we are still working." Parents were skilled at providing excellent care. They became proficient at giving chest physiotherapy and postural drainage, providing rehabilitation therapy, changing dressings, caring for feeding tubes, using equipment such as suction apparatus and feeding pumps, or managing the multiple drug regimes the child required. At the beginning of the child's illness, parents were unsure what to do. As they became familiar with therapies, they gained more confidence. As one mother said, "You get used to it. You get confidence to do it. Now we know what to do."

Parents provided care that became increasingly complex as the child's illness deteriorated. Some parents learned how to provide care simply by watching professionals as they worked. In most cases, hospital professionals formally taught parents the skills required. Professionals did not always recognize, however, that parents needed time to learn the skills. Sometimes, families were discharged from hospital with minimal preparation and parents felt incapable of performing tasks. Even with a week of training in the hospital, it often took two or three months before parents were comfortable being alone and having sole responsibility for the child's care.

The child's reduced ability to communicate made care provision more difficult. Verbal communication is usually important in everyday interactions, but children with NLTIs were unable to converse once their illness had taken hold. Parents had to learn or create other ways of communicating with the child, although they still continued to talk to the child. They often reverted to the types of communication they used with pre-verbal infants and young children. Parents looked for subtle clues that the child needed attention. They became adept at interpreting the child's body language. One father noted that the noises his son made could mean that he's happy in one situation, but unhappy in another. The father said that he became "attuned to it" and could tell what his son wanted "just by the expressions on his face." As one mother explained:

She won't talk, but she moans and groans. And you just don't have a clue what she's moaning and groaning about. But because I'm her mother, and because Adam too [her father], we both know, just know. Either her diaper has to be changed or she wants to be fed or she wants to be walking or she wants to have a music video on or a book read or something like that, or [go] outside.

Tactile communication often increased to balance the lack of verbal communication. Body-to-body contact, such as sitting in a chair holding the child, and other forms of physical contact, such as kisses and hugs, became important vehicles of communication.

Parents were very creative in finding ways to provide the child's care. They often faced situations where the child's condition prevented the use of standard procedures or typical equipment. One father explained:

We find we have to be very creative with ideas. Unfortunately, no doctor or anybody else can really come in and say, "This is what you should be doing." It is something that you learn from experience. You find that you have to be creative.... Sometimes, you don't have everything and you say, "Okay. Fine. This is all we've got - let's make it work."

Many parents learned through trial and error or their own ingenuity. For example, one child had a habit of biting down on a glass. His mother then ensured that he only drank through a straw so he would not injure himself. Parents adapted their furniture and equipment to make life easier. One mother used plastic lawn chairs for her son to sit in. They made it easier for her to "move him in and out, plus he can't fall." Another family put their son in a big bean bag chair to keep him safe. One family commissioned special chairs for their children. The multi-coloured chairs were on wheels and could be tilted. They were also well padded and supported. The mother believed that conventional wheelchairs do not provide support for a long period of time. She wanted to ensure that her girls were able to be up out of bed for a reasonable length of time and still be comfortable. Many of the children vomited frequently. One family devised a way of reducing the mess and their stress. They "just grab a receiving blanket. She throws up in it, we chuck it and we grab another one. So we go through a lot of those, just wash them."

Another family found that the weight of their son's bedclothes made his foot drop worse. They improvised with a shoe rack to keep the bedding away from the child's body. This family also used a hammock when the child was out of bed to rock him gently and to keep him safe. Safety was a major concern for most families. Many of their innovative ideas were designed to ensure safe care. In one family, the parents designed a safe area with large plastic covered cushions on the floor and plastic covered side barriers that were a few inches high. The parents had the cushions specially made. When they acquired a second set, they then gave the old ones to

the child's school. Professionals were usually impressed by a family's ideas, and they sometimes shared the ideas with colleagues.

Much of parents' time was taken up in providing physical care to the child. Despite some respite, most families provided the majority of care to their child. Parents learned ways of managing the physical care from professionals. But parents also were creative and devised new ways to provide care in difficult circumstances and to teach professionals.

Promoting the child's health

The primary goal for many families was to keep the child as healthy as possible. Parents hoped for two resulting benefits: to keep the child alive longer, and to lessen disruptions to family life. Parents worked hard keeping children safe, preventing them from becoming ill, and providing them with good nutrition. They continually monitored the children to identify potential threats to health.

Feeding the child was a priority for all families. None of the children could eat regular food and most of them required feeds through a naso-gastric or gastrostomy tube because of feeding disorders related to their illnesses. Many of them suffered from gastric reflux so the feeds had to be given slowly with the child's head elevated. Parents emphasized the importance of adequate nutritional intake. Some parents insisted on their own "home made " meals that they cut up or pureed to ensure optimal nutrition. Many equated feeding with life and strove to maintain the child's life. "Our child was dying. We had no choice. She wasn't eating. She needed the tube."

Emphasis was placed on increasing a child's weight or at least maintaining it. Families went to what seemed almost extraordinary lengths to ensure that the child received adequate nutrition. In one family, the child received feeds overnight, because both parents and professionals were worried that he was not getting enough calories to sustain him and to help him grow. Each day, he received about two cans of liquid, commercially-prepared food for

children. His parents also gave him additional water to increase fluids. They diluted each can of food with 100 ml of water because it allowed the feeding to flow. They also flushed the delivery system with water and a readily available soft drink to ensure the tubing remained unclogged. The child had an intake of about 500 calories per day. He had a gastric reflux problem which made him susceptible to aspirating his liquid feeds into his lungs. He received about 50-60 ml/hour continuously during the day, and 20-30 ml/hour at night, although it might run faster when a nurse was present. Parents continually monitored the feedings and slowed down the rate at the slightest sign of reflux.

In another family, parents gave their daughter one ounce every 15 minutes, signalled by a timer, over about one and a half hours. They fed the child through a syringe into her gastrostomy tube. In school, the food could not be given that way because of the time it took, so they used an electric pump to infuse the food continuously. Neither of these families were exceptional among the families interviewed. Feeding took priority over everything else and the family's routine revolved around the feeding schedule.

Families lived in fear of the child developing pneumonia or another illness that might hasten the child's death. They developed strategies to limit contact with others who might be infectious. If the child attended school and other students were ill, then the child was kept home until the perceived danger was over. Nurses and other workers in the home had "strict instructions that if they have, if they think they are even coming down with anything, don't bother setting foot in the door." Parents with even relatively mild illnesses such as colds did not provide care to the child wherever possible. The other parent assumed full care, or, "You wear rubber gloves. You don't breathe anywhere near her trying to keep your face away. You wash your hands 500 million times in a day."

Families also stressed the importance of helping the child maintain mobility as much as possible because "the greatest health problems start setting in when you're not mobile." They

hoped to keep the child alive as long as possible so they worked hard at maintaining or improving the child's physical and cognitive skills.

Parents exerted intense effort to mobilize the child earlier on in the illness when the child had the most motor skills. For example, families took the child horse-back riding to improve balance and muscle strength. As the child became weaker, strategies changed so that a walking frame became more important. Then, when the child could no longer walk, a standing frame was used. Most parents only realized that efforts at rehabilitation were no longer working when the child's mobility was severely restricted: the child spent most time in a wheelchair or was confined to bed. At that point, most parents stopped aggressive exercises. They continued to encourage the child to move in whatever way was possible, even if just moving slightly to music. Most of these children enjoyed visual stimulation - television and movie videos were favourite past-times. The children also often enjoyed taped stories and music and many had their own personal stereos. In most homes, the television was on whenever the child was awake, even though parents did not like television.

Parents worked diligently to promote their child's health and prevent illness. They wanted the child to live as long as possible. So, they took measures to ensure the child maintained optimal wellness. Keeping the child safe, preventing illness, and providing good nutrition were important to parents. The child's optimal wellness also minimized disruptions in the family life.

Forming New Relationships

As previously discussed, extended family and friends often withdrew over time and most families also withdrew from others. Parents had limited time and energy to spend on other people. In addition, parents found they had little in common with families where all the children were healthy. So, many families made new friends with other families who had a disabled child, or they joined an organization for families whose children had the same disease. One mother likened dealing with people whose children had a debilitating illness to a "safety net," because

they were "all in the same world." Families found it easier to talk with other families of disabled children. They received support and understanding from other families who understood because they were going through similar experiences. This father said:

They always do say as well that birds of a feather flock together. So, because we have a child that is handicapped, we are finding now that we are probably more...friendlier, we are able to adapt better and communicate better with families that are going through the same crisis, I guess. It need not have to be the same type of disease. Like the one time we went to the M. R. Foundation [name of disease]. They had a Christmas party and there were other families there. We were able to relate to them and that felt good.

Parents gained information from other parents and organizations. They also shared what they had learned with others. Most parents benefited from this mutual sharing and contact. However, belonging to an organization for dying children also had its disadvantages. Families became friendly with other families whose child later died from the disease. Parents found it very difficult when another child died. They were forced to think about their own child's prognosis. A few parents limited contact with other families of disabled children or organizations because they could not face the prospect of their own child's death. One mother voiced her ambivalence in this way:

Something else that is different is we have been thrown into a whole different world of wheelchairs and other children with disabilities. I think part of it is good, but there is so much that is not good. I'm just thinking about R.M. because we got the call, I got the call yesterday [about a child's death]. That is one of the things that I hate the most....If you were isolated, if you didn't belong to an organization like this, it would be very difficult because nobody understands how you feel. But there comes a price with us being involved. Now, you grieve for other children and other families. It just brings it home to you. I don't know. Sometimes I question whether that price is worth it. I guess it is, I guess it is. You have to have contact with other families, but sometimes you get so sick and tired of, like this is what your life revolves around, and you find it very difficult to talk to your normal friends.

New relationships with other families of disabled children helped fill a gap in families' lives. Families gained information, support, and strength to carry on caring for their child. However, contact with families where a child died from the illness was also difficult because it was a continual reminder of their own child's mortality.

In summary, *holding the fort* characterized families' experiences as they gained some control over the situation. They spent most of their lives on plateaus that were fragile and tentative. Their view of time changed in their efforts to manage the work accompanying their experience. Parents spent a lot of time and effort uncovering and organizing services for their child. As the child's condition deteriorated, care became more complex and services increased. Families became more dependent on professional caregiving. Subsequently, they faced issues, such as lack of privacy, that arose because of increased numbers of caregivers in the home. Parents learned how to provide physical care to the child and they strove relentlessly to maintain or improve the child's health. Finally, families formed new relationships with other families of disabled children because they gained support from such families. The downside was that the death of another child raised the spectre of their own child's prognosis.

Subsequent Precipitating Events

Navigating uncharted territory was a continual process that was originally set in motion by the initial precipitating event. Over time, families shifted their priorities, created meaning out of the situation, and settled into a period of relative stability. Although families learned about their new territory and found ways of managing their new world, that world did not stay the same for long. Subsequent precipitating events, such as a decline in the child's physical health or a need for new equipment, sent families into unfamiliar territory once again and impacted on the other dimensions of the process. Families described this time as *dropping off the plateau* (see Figure 2).

Dropping off the Plateau

Families adjusted their lives every time their child's condition changed. In most families, changes were most rapid at the beginning of the illness. Changes eventually slowed down, however, there was no predictable pattern for families. Occasionally, a child's condition even

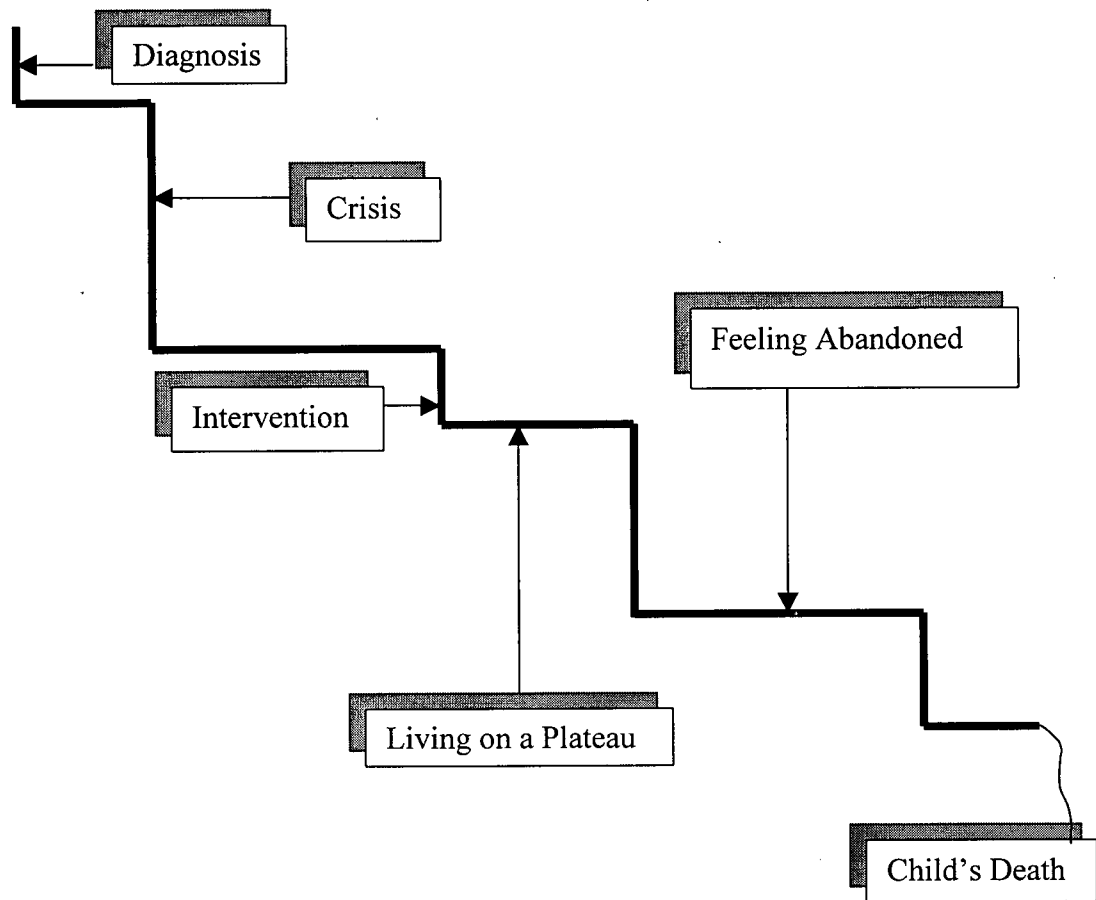


Figure 2. Dropping off the plateau

improved slightly for a period of time. Yet, parents knew that the child was enmeshed in a downward trajectory towards inevitable death.

The trajectory, however, was not a steady decline. Instead, families lived their lives on plateaus of relative stability, waiting for the next crisis or sudden decline in their child's condition. While on the plateau, most parents' thoughts about the child's illness and prognosis remained in the background. When the child became really sick, parents thought more about the illness and prognosis and these thoughts moved to the forefront. During each crisis, families dropped off the current plateau and fell until they reached the next plateau:

She will go along on a plateau of doing things. Well, say for instance, like right now she is going along seeing things and then all of a sudden she will drop off not seeing anything.

The extent of the drop depended on both the severity of the precipitating event and the available corrective options. In the beginning, families were often faced with short plateaus accompanied by sharp drops as the child lost abilities. As the child's condition worsened and there was little more to lose, then the drops usually became less severe. Treatments could shorten the drop, but treatment options became more limited as the child's illness progressed. Families also worked hard to extend the length of the plateaus. They expended much time and energy in promoting the child's health in order to prevent a deterioration in condition that would push them off the plateau.

Families spent most of their lives on plateaus. Life on a plateau was easier than dealing with crises, "Right now we are at a stage where he's reached a plateau. Although he is still deteriorating, we can cope because he is moving at a really slow pace." Yet, many families also felt alone and frightened on the plateaus. They received little help and felt abandoned by the health care system. They often interacted with and received help from the health care system when they were in crisis, but they had little contact at any other time. Professionals seldom recognized a family's need for ongoing support when the child's condition was relatively stable.

Dropping off the plateau affected all other dimensions of the process. Fear, uncertainty, or grief increased, depending on the particular precipitating event. Each time families fell to the next plateau, they faced new territory again. Consequently, they needed to learn about this new area. They needed to find ways to manage their world once again. So they sought more information in order to manage the associated work. Families often had to shift priorities again, such as increasing their focus on the child. Some families determined that they had even less time to deal with other people, so they got rid of more excess baggage. Finding meaning in the experience required a further examination of their situation. In most cases, families settled at a different level of holding the fort as they added other services to their routine or learned how to provide a new treatment. Families continued moving through the process of *navigating uncharted territory* as they encountered further precipitating events. Eventually, the process would end with the child's death.

Intervening Conditions

The strategies that families used to manage the phenomenon of *navigating uncharted territory* have been described above. However, these strategies were influenced by intervening conditions that reflected the broader structural context of the phenomenon. Four intervening conditions, *relationships with health care providers*, *availability of information*, *gender differences*, and *communication between parents*, affected the course of the illness experience. Each condition facilitated or constrained the strategies that families were able to use. The four intervening conditions will be described in the following section.

Relationships With Health Care Providers

Health care providers greatly influenced families' illness experiences and could facilitate or inhibit the extent to which families were able to successfully navigate uncharted territory. In most cases, a physician was the pivotal figure in families' interactions with the health care system. Physicians traditionally are invested with decision making authority as senior members

of the health care team, and they also function as the entry point to the system. Therefore, parents expected this pivotal role. In addition, families interacted with many other health care professionals, such as nurses, social workers, physical therapists, and home support staff. All of the relationships were dynamic and evolving over time.

Parents also emphasized the importance of an ongoing relationship with a few key individuals. Since the health care system is focused mainly on providing care to individuals who are ill, families often only met with health care providers when their child was sick and in crisis. Yet, families spent most of their lives at home, so professionals failed to see the whole picture of the families' lives. Some professionals then made decisions based on these intermittent, crisis-induced interactions, decisions that were often irrelevant to the families' everyday lives. For example, a dietician insisted that a child needed a gastrostomy tube because of his low weight, but he had lost weight because of an episode of pneumonia. As his father said:

They're looking at this situation right now, right here, right now. But it's like right here, right now is diddly on the overall perspective. It's like this nutritionist. She only sees him when he's sick, so she wants a [feeding] tube in him. She's never seen him when he's in good health. Or on a good day.

If their contact with a particular provider was brief, then parents merely viewed the person as an extension of the health care system. However, where individuals had intimate involvement in the ongoing management of the child's illness, then the relationships with those health care providers became very significant in the families' lives. Despite the parents' perceptions that relationships were extremely important, parents suggested that, in many cases, health care providers were insensitive to the impact of their discourse on families. When parents were dissatisfied with their encounters with health care providers, such as when they perceived the professional's care was incompetent, then relationships became strained and negatively influenced a family's ability to manage the child's illness.

Most families found that their views about health care providers changed as the illness experience evolved. At first, many parents assumed that health care providers would understand their concerns and would try to find a solution. In addition, parents had complete trust and confidence that professionals could and would solve all health problems. Moreover, parents expected health care providers to know what was best for the family. They accepted professionals' initial answers and did not question them. Inevitably though, this trusting and uninformed position was insufficient to meet the families' needs for answers to their concerns.

For some families, a single event shattered their trust, while for others, a series of experiences gradually challenged their trusting and uninformed stance. Some families believed that professionals deliberately withheld information. In other cases, parents perceived that the health care providers' overall objectives for the child's care were so different from their own that the relationship was no longer tenable. For example, during one child's acute illness episode, a physician advised her parents to "just let her" die. Yet, the parents believed that the child could still enjoy a good quality of life.

When trust in health care professionals was shattered, parents felt as if they were in a nightmare. They could no longer trust that their child would receive adequate and competent care. It was a time of anger, anxiety, and frustration. Some parents were filled with self-doubt. Their perception that they had made bad decisions in the past when they had trusted professionals triggered doubts about their future ability to make good decisions about their child's care. Moreover, some parents also questioned their ability to make any important decisions. This father said:

The whole thing of trusting the hospital environment to do what they were doing to my son. The way we were treated, and the way we were disposed of, has really left me feeling incapable to make any other decisions of life importance. I feel like I made a totally wrong decision by letting those wackos near my son. I'm just afraid of making any other decisions along that line. For myself, or him, or my wife. I'll make the wrong decision again. It's not worth it.

Some parents reacted with anger and hostility towards health care providers they no longer trusted. They worried, however, about the impact their actions might have on the child's care. In at least one instance, a professional refused to treat the child in retaliation to the parents' assertive stance.

Feelings associated with distrust of professionals were very unpleasant. Parents could not remain extremely angry and anxious for an extended period of time. Moreover, parents recognized that their child needed health care. Therefore, parents needed to find ways to re-establish limited trust while still maintaining some skepticism about the health care system. The realization that they themselves were capable of being advocates for their child and that they could make choices freed parents to think in different ways about the health care system. Parents started to understand how their family fit into the larger context of health care. They recognized the constraints in the system and realized its limitations. As one father said:

They suck you in at the beginning that you're going to be an important part of this team. And you're fuck all. You're nothing. They don't give a shit what you think or what you want. Anybody that's an important part of the team, I can't see an important person sleeping on the floor. But that's where you sleep as a parent. You sleep on the floor. Does the doctor sleep on the floor? Does the patient? No. That kind of thing just pissed me off.

Consequently, parents changed their attitudes about their own responsibilities. They learned that they needed to take charge of the situation. Parents realized that they had important competencies themselves. Moreover, they were the experts on their own family's situation. They affirmed the expert knowledge owned by health care providers, but they valued their own expert knowledge as a distinct entity. Parents needed to develop confidence in their abilities. They also needed advocacy skills and to be assertive and confrontational in some health care relationships. Unfortunately, despite the promise of family-centred care where families were considered part of the health care team, some health care providers continued to dislike co-operative models of care and were reluctant to involve parents in caring for the child.

Parents gained confidence through a variety of strategies. Just as poor decision making in the past induced self-doubt, so success in health care decision making increased parents' confidence. Parents also mobilized external resources, such as having multiple health care relationships rather than depending on one person, or asking for a second opinion. In many cases, parents learned how to access and use medical libraries and the internet. They taught themselves how to read and evaluate research reports. They also contacted international experts on the child's condition and attended conferences to learn as much as they could. Lay support groups and other families with a child who had a similar condition provided sources of support and taught parents how to improve their advocacy skills.

However, acquiring a sophisticated knowledge of the child's illness and its management, and accepting major responsibility for the child's care were not easy tasks. They required an extensive time commitment, dedication to learning, and access to resources for developing the knowledge. Parents generally shared the task, but in most families one parent, usually the mother, took on the major role. In one family where the father also stayed at home with the child, the father took charge of this role. The time and effort involved in keeping current and taking the major responsibility for the child's care were not always recognized by health care providers, because, as parents commented, much of the work occurred "behind the scenes."

Once parents no longer either blindly trusted or completely distrusted every individual in the health care system, they became highly selective in allocating their trust. Each individual professional had to earn that trust. A parent's trust in a professional was dependent on the professional's ability to trust the parent. The relationship could not be optimal unless the trust was reciprocal. Professionals demonstrated trust by accepting parents' knowledge and expertise. Too often, especially in hospitals, professionals did not listen to parents. A few parents felt that they almost had to explode in rage before anyone would listen to them. One father said sadly:

Behind it all, you could tell deeply that everybody's really trying to help. But it becomes systematic and regimented almost in the hospitals. "This is the way we do it and this is when we do it. Whether you understand it or not, it's just part of the flow that's going to happen." So what that forced, for myself anyway, was that everything had to get right to the point where you're ready to explode before there was any real communication....It seemed that you had to, I had to anyway, build up to the point where you're ready to explode or almost lash out at somebody [before anybody listened].

Parents changed physicians, fired home care nurses, or asked for home housekeeping workers to be replaced if, in the parent's view, the individual was not trustworthy. Competence, as reflected in technical skills and strong clinical reasoning, was a prerequisite to trustworthiness. But, competence alone was not sufficient. Competent health care providers also needed to possess good communication skills if parents were to consider them trustworthy. Communication reflected active listening skills, giving clear and understandable explanations, and being willing to talk about what day-to-day life was really like for the family.

Parents highly valued health care providers who respected each member of the family as a unique and competent individual. Willingness to reveal something about their personal lives and to act informally were health care providers' attributes that parents also appreciated. Finally, professionals needed to be willing to learn and had to be accessible when families needed their assistance. Lamentably, parents often had difficulty in finding a suitable health care professional, because they considered that trustworthy professionals were atypical and were different from the norm in health care situations. However, there were some health care providers, typically among those providing care in the home, who were deemed highly trustworthy by parents and they became "just like a part of the family."

Occasionally, an initial health care provider, such as the physician who diagnosed the child's illness, actively encouraged parents to have confidence in their own knowledge and abilities. These early interactions between families and health care professionals provided positive role models and set up expectations for future relationships. A satisfying and collaborative relationship was implemented from the beginning of the child's illness.

Professionals involved admitted that they did not have the answers and that they were learning as they went along. They encouraged parents to ask questions and to build up their own knowledge. Parents who had open and honest communication with professionals reported that their interactions were relatively trouble-free. One mother said that she had always treated the physicians "as consultants, rather than as doctors." She was the person in control of her child's care, not the physician. She acknowledged, however, that she was a very vocal parent for her child's needs and her style may have influenced the relationships.

Compared with most parents, those who were treated as knowledgeable partners from early on in the illness experience described fewer feelings of anger and frustration with their primary health care providers. They felt more in control of the situation. In addition, they were confident and assertive in their dealings with other health care professionals. However, these parents also reported some instances when they could not trust a health care provider. In such cases, they used a standard for trustworthiness similar to that of the other parents - a combination of competence and communication skills - to judge the health care providers.

Some parents developed strategies to work with health care providers in ways that increased their chances of receiving sufficient and appropriate care. They learned how to "manipulate the system" to their advantage. These parents focused not on the relationship as such, but concentrated on the services they needed and figured out ways to obtain the services. For example, when they needed increased help in the home, these parents learned that they were more likely to receive that help if they told the social worker about their "worst day." Some of the strategies were learned from other parents who had dealt with the system. Many times, parents simply learned what worked and did not work through "trial and error." When parents "got smart and went around the wall," they were more satisfied with their experience.

Availability of Information

Developing adequate knowledge and obtaining information related to the child's illness were strategies that all families reported as helpful in their struggle to live with their child's illness. In the months or even years prior to diagnosis, families frantically sought out information in an effort to understand what was happening. Once the child's disease was identified, families then tried to learn as much as they could about the medical treatment and prognosis. They wanted to learn how to provide the best care for their child. Knowledge also helped to reduce their fears and uncertainties. It gave them some control over the situation.

However, in most cases, little or no information about the illness was available. Neurodegenerative diseases are uncommon, so, professionals often had little information about the trajectory of the illness. Parents were frustrated when they could not find the details they needed. In some cases, they initially suspected that the health care providers were withholding information. However, parents realized over time that there was a paucity of documented information about neurodegenerative illnesses. Despite strenuous efforts to obtain information, parents were often left "stumbling in the dark." They had little guidance on their journey. The lack of information often increased their fears and uncertainties. Typically, availability of information about the particular illness was a constraining factor for these families.

Gender Differences

Parents did not necessarily move through the process of *navigating uncharted territory* in tandem. Gender differences sometimes led to conflict and misunderstanding between parents and posed a threat to the survival of the family. When parents were unable to resolve these differences, the marital unit was severely strained. However, when parents accepted the differences and understood that it was alright to have differences, then the tensions were reduced and the family became more stable. There were often differences in how mothers and fathers

grieved; in how they responded to medication and feeding regimes; in the amount of information they required; and in how well they cared for themselves.

Grieving.

Both mothers and fathers were deeply affected by the child's illness, but fathers' grieving was often done privately and was not as open as mothers' grieving. It was not always obvious to others, including spouses and professionals, that fathers were suffering as much as mothers. Most mothers and fathers recognized that they grieved differently. Mothers were more emotional and verbal, whereas fathers tended to withdraw and think about things quietly by themselves. While mothers wanted to talk, fathers felt little need to talk. Instead, they wanted "action." Fathers took each day as it came and "got on with it," neither dwelling on the past nor looking into the future. These differences sometimes caused marital conflict as suggested by this mother:

The women want to talk about it and the men just keep pushing the women away and saying, "No, no, no, I don't want to talk about it." And then it causes conflict. So that's probably a lot of the reason that a lot of couples that start off together are not together as it [child's illness] progresses....Fathers usually don't really want to talk about it much....I don't know what it is, that male gene that just says, "I know it's happening, but that's it. I don't need to talk about it. I know everything there is to know, I don't need to talk about it." Where a woman, it makes you feel better when you talk about it....It was like I NEED to talk about it and he needed NOT to talk about it, so it started fights and conflicts.

Some fathers suggested that talking about the situation was not helpful to them. Instead, talking only invoked thoughts about the future that detracted from their ability to deal with the present.

As this father explained:

We've gone to counselling. I guess it's helped her. But I don't think it's helped me at all....I don't really need the help. I take it day by day. [If] you look at the whole picture and [talk about] "Where am I going from here?" it would drive you nuts. No, you can't look at it like that.

Mothers found the different ways of grieving very hard to accept. In reply to the interviewer's comment that people sometimes get angry with one another because they express emotions differently, this mother said tearfully:

I do. I don't understand why I never see a tear in his eye or anything like that. Where me, I can just cry so easy and he doesn't. And I don't understand. Doesn't it kill him? Do you know what I mean? Doesn't it hurt him like it hurts me?...I just find it hard that he doesn't show any emotion. He doesn't. Where me, it just kills at me, kills. Just hurts me so much inside. Where[as] I can look at him and he doesn't even show any [emotion].

As a consequence, her husband said, "She gets really...unco-operative with me when she realizes that I don't grieve the same way she does."

While fathers agreed that they grieved differently from their partners, they did not necessarily agree that the differences were due to gender. When asked about the reasons for these differences, one father said:

I think we are both two very different people, and I think we both react very, very differently to given situations. I honestly think that my background and my make up enable me to just deal with what's there. This is what I have to do, and so I do it....I think that we're both lucky that my make up enables me just to keep going and deal with what has to be done....I would tend to say that I'm more rational in the way I deal with things, and she's more emotional in the way that she deals with things.

Medication and feeding regimes.

Mothers and fathers often disagreed on the need to follow a strict regime for giving medications or feeds, especially the naso-gastric or gastrostomy tube feeds most of these children received. Mothers ensured that medications and feeds were given as close to the prescribed time as possible, whereas fathers were much more relaxed about the need for a strict schedule. Even when the father asked a professional to explain that the schedule could be flexible without the child being harmed, the mother usually remained firm that the medications or feeds needed to be given at set times. Fathers became exasperated if mothers remained inflexible and refused to adjust the timing of the child's medication or feeds to fit better with the family schedule. One father explained his point of view:

She's just regimented over this schedule. Like I said, I try to plan what will happen when as well as I can so that everything's fairly in order. But I live by that "Well if we're 15 or 20 minutes off, we're 15 or 20 minutes off which is not going to ruin the day."...I don't know, maybe it comes down to this maternal thing of they feel like they gave birth to the child and they don't want to do any more damage....Yes, it would be ideal, in the ideal world where the child gets his medicine on time every day. But if all of a sudden a rock

goes through your window, or you spill a pot of coffee, or baby spits up or whatever, [then] things get jumbled around and we adjust and it's not the end of the world.

Sometimes strictly adhering to a schedule caused difficulties in taking the child away from the home. As one father noted:

She still won't think and say, "Well, maybe I just won't feed them." I mean, every kid misses a meal, but Karen is programmed and she runs on program....[She'll say] "Well, we'll be out past 2 o'clock, so they must have a feed there." And [she] won't give any consideration to, if we maybe go to a mall or maybe go to a movie, maybe just don't feed them. And it's not saying don't feed them, but feed them when we get home. Just don't have it so blatantly, with the beep, beep, beep [of the feeding pump] in the middle of the movie. Everyone turns and looks. So, I don't think that has sunk into Karen.

The arguments caused by these differences in attitudes were often ongoing and of long duration. Over time, this source of conflict became a major issue in some families.

Information needs.

All parents sought information in order to alleviate their emotional distress, and to increase their knowledge about the child's illness and its management. However, mothers generally had the more intense need for information. In many cases, mothers continued to search for answers to questions, such as what had caused the illness or what could be done, long after fathers had stopped asking such questions. Mothers often spent time and energy finding information that they then shared with the fathers. In addition, some mothers were particularly interested in information about the genetic aspect of the illness. A number of these diseases were transferred through maternal genes, and sometimes a mother felt guilty and responsible for causing her child's illness.

Caring for self.

Because the focus of attention was on the child, parents tended to neglect their own needs. For example, time for physical exercise or preparing a nutritious meal was at a premium. Mothers, in particular, perceived their own needs and comfort as an "add-on" rather than integral to their own health. Once the needs of others were met, then mothers might take care of

themselves. Fathers were more likely to take care of themselves. For example, they would incorporate exercise into their daily schedule. Sometimes fathers tried to encourage their partners to exercise, but they met with resistance that frustrated them. While both parents recognized that they needed to be healthy in order to care for their child, mothers had difficulty in taking time for themselves.

Communication Between Parents

Another condition that influenced the strategies used by families was the extent to which parents were able to communicate with and understand one another. In all families where both parents were involved with the child, parents acknowledged marital difficulties. Parents had no time for each other. They were so busy concentrating on the ill child that they neglected each other and their marriage. Yet, the marriage was the glue that held the family together. Having a strong marriage before the child was diagnosed contributed to parents' abilities to weather the difficulties they experienced. Even strong marriages were severely shaken by this added stress and required effort to remain intact.

Communication was the central key in those families where parents were most successful in making their marriage work. However, successful communication did not happen easily, even in families where communication had always been very good before the child's diagnosis. Instead, successful communication was only achieved after months of arguments or misunderstandings. Parents noted that although it was hard to find time to talk with one another, they needed to talk. Understanding their partner's point of view was essential if they wanted to maintain their marriage. Although parents recognized that communication was important, they usually found that their existing communication skills were inadequate for their current situation. Even parents who had excellent communication skills before the child's diagnosis were initially unable to find the words they needed. Parents had never been in such a situation before. They often needed external assistance to help improve communication within their marriage. Often,

parents had forgotten how to talk with one another. In many instances, they were upset because they did not understand why their partner's reaction to the situation was so different from their own. Discrepant views were frequently a source of conflict. When parents accepted differences, then discrepant views could also be a source of strength if the different perspectives were viewed as complementing one another.

Parents used multiple strategies to enhance both opportunities to talk and communication skills. They recognized that they needed time together if they were going to be able to talk. However, finding that time often meant that they had to trade off something else, such as sleep. Respite was also very important to families as it provided time for parents to talk. This vital benefit of respite was not always recognized by professionals. Not all families were able to take full advantage of respite time. Lower income families lacked the money to pay for sufficient respite care. Families with higher incomes acknowledged that they could "have more fun," because they could afford to "play a little bit more than other families," such as going out to a nice restaurant while their child was receiving respite care. Those families sometimes felt guilty about accepting respite care that was paid for by the government or a charity. Yet, they strongly believed that the respite kept their family together.

The effort involved in going out for an evening or going away for vacation also deterred parents who were already exhausted. They had to arrange for child care, and the local teenager or grandma was usually not capable of caring for a child with complex needs. Parents were restricted in who they could ask to baby-sit. They were often dependent on professional caregivers who were not always available and whose services were quite expensive. Despite the benefits of respite care, going out together was not always worth the effort.

Learning how to improve communication skills was another strategy used in some families. In one family, parents took a course on chronically ill children that helped them tremendously. "It got us talking again...and gave us a better understanding of where we each

were in the stream of coping." Some parents found that counselling enabled them to express their emotions and helped them to learn how to listen to their partners and to understand the other partner's reactions. Counselling was not successful for all families. In those families, parents continued to have marital difficulties.

Summary

Families moved through a process of *navigating uncharted territory* as they lived with a child who was dying from a neurodegenerative, life-threatening illness. This process was initiated by a precipitating event, such as one parent recognizing a deterioration in the child's motor skills. The emotions of fear, uncertainty, and grief gave impetus to the process. These emotions were very strong in the beginning. They gradually declined over time, but increased with subsequent precipitating events. The process of *navigating uncharted territory* was characterized by four dimensions - *entering unfamiliar territory, shifting priorities, creating meaning, and holding the fort*. Parents used strategies within these dimensions to manage the physical, cognitive, and emotional work arising from the situation. These dimensions and strategies were constrained and facilitated by four intervening conditions - *relationships with health care providers, availability of information, gender differences, and communication between parents*. In addition, the process of *navigating uncharted territory* occurred within the broader context of the *acute, curative health care system* and the *sociocultural environment*.

Chapter VI: Discussion

In this study, families moved through a process of *navigating uncharted territory*. This process and its dimensions have not been previously identified in the literature. The findings presented in the preceding chapter provided answers to the research questions that were posed at the outset of this study. Families' perceptions and experiences of living with a child who has a neurodegenerative, life-threatening illness (NLTI), and how those perceptions and experiences changed over time, were documented throughout the process. The impacts on families of living with a child who has an NLTI were profound. These impacts were highlighted by the work that families undertook. Finally, the factors that families perceived enhanced or undermined their ability to care for their child were described within the context of the experience and the intervening conditions. The purpose of this chapter is to place the findings of this newly developed grounded theory within the broader context of current research and literature. First, the process of *navigating uncharted territory* will be compared and contrasted with two of the existing stress, coping, and adaptation frameworks. Second, the nature and shape of the illness trajectory will be discussed. Third, discussion will centre around the concept of family-centred care and the influence of health care relationships. Finally, key themes about the toll that the experience takes on families will be identified and discussed.

Stress, Coping, and Adaptation Frameworks

Existing stress, coping, and adaptation frameworks provide some direction for education, research, and practice. However, these frameworks restrict one's view of families' experiences to only those aspects that involve coping strategies. Further, in coping frameworks, problem-solving coping strategies are often perceived as more desirable than emotional or cognitive strategies, and cognitive strategies are usually associated with passivity rather than activity (Morse & Fife, 1998). There is, however, growing recognition of the importance of cognitive strategies when a situation cannot be changed or a problem is insoluble (Morse & Fife; Nolan,

Grant, & Keady, 1996). A major dimension in the process of *navigating uncharted territory* was *creating meaning*. Cognitive strategies that allowed families to create meaning in their situations, such as taking one day at a time, seeing the funny side of the situation, realizing there is always someone worse off than yourself, looking for the positive things in the situation, or drawing on religious beliefs, were important because they helped families manage their perceptions of the situation. Rather than being passive strategies, these cognitive coping strategies reflected active, creative, and resourceful ways of managing difficult situations. They were characterized by a diversity and pervasiveness that has seldom been acknowledged in the literature.

Recent research provides some support for the usefulness of cognitive strategies in coping with circumstances that cannot be changed. Clarke-Steffen (1997) developed a model of the family transition to living with childhood cancer in her grounded theory study. A major process that these families used was assigning meaning to the illness. This process encompassed some of the cognitive strategies used by families whose child had an NLTI. In her research with families of children with cystic fibrosis, Bluebond-Langner (1996) also identified cognitive strategies that families used to create meaning. For those families, the aim of many of their strategies was redefining their view of normal so they could continue to view their own family as normal. Family caregivers of mostly older people have also indicated that they found 11 out of 15 possible cognitive strategies helpful (Nolan et al., 1996). Most of these strategies were similar to those used by families in this current study to create meaning. It appears that in circumstances that are not amenable to change, such as when a child has an NLTI, the importance of cognitive strategies should not be undervalued. Instead, the use of helpful cognitive strategies should be encouraged and viewed as an important addition to a family's repertoire of coping strategies.

Individual Coping

As previously discussed in Chapter II, the Lazarus and Folkman model (1984) has limited applicability in research with families living with a child who has an NLTI. One cannot explore

the family's perspective within a model that relies on the individual's perspective. In addition, coping is only one part of a family's life and is in response to chronic rather than discrete, situational stressors. Further, the subconscious nature of coping efforts that are routinized impedes one's ability to uncover strategies that are used on a daily basis. Recent theorists and researchers have recognized that coping with chronic stress has been understudied (Gottlieb, 1997). Little is known about the behavioural and emotional regulatory processes that people use in the face of persistent life strains. Gottlieb noted that the conceptualization of coping in the face of ongoing adversity is broader than the definition used in the Lazarus and Folkman process coping model. In more recent approaches, coping is seen as part of the "ongoing lifecourse process of adapting and accommodating to transitions, discontinuities, and other destabilizing and threatening experiences" (Gottlieb, p.4). Many conditions fall under the umbrella of chronic stressors, one of which is chronic illness.

There have been many research studies with individuals facing chronic illness. However, as Gottlieb (1997) pointed out, researchers typically ask questions about coping efforts at times of acute stress, such as might occur when a child with an NLTI develops pneumonia. Gottlieb questioned why coping efforts that are aimed at acute episodes should be viewed as representative of an individual's life experience. Instead, he continued, coping during an acute episode of illness may be different than coping when the condition is stable. Moreover, multiple and interacting stressors may be common throughout the period of chronic hardship. The dynamic relationships between time-limited stressors that occur only once, recurrent time-limited stressors, and ongoing, open-ended stressors have not been explored (Gottlieb).

Results from this current study provide support for Gottlieb's propositions (1997). Families were faced with multiple potential stressors throughout the process of *navigating uncharted territory*. They needed to complete physical, cognitive, and emotional work in order to manage the many different aspects of the experience. In addition, families used a variety of

coping strategies that changed depending on whether they faced crises or were living on a stable plateau. Moreover, some of the strategies had become part of a family's life and were no longer recognized by families as coping strategies. The researcher, then, had to link families' stories with the chronic background of life with a child who has an NLTII. This conceptual approach has implications for how data are collected in chronic illness. For example, checklists that ask respondents to identify coping strategies they intentionally use to manage specific stressful demands would likely fail to capture the range of coping used in chronic illness. Other tools, such as in-depth interviews or daily diaries, that can address broader issues may be required instead.

Some researchers have begun to investigate the various conceptions and contexts of chronic stress from this new perspective. They are exploring the responses that individuals make when faced with prolonged exposure to stressors and difficulties in life. Much of this research is occurring in the fields of psychology and sociology and is not focused on chronic illness per se. One chronic illness study (Folkman, Moskowitz, Ozer, & Park, 1997), however, was reoriented to this perspective when researchers realized the limitations of focusing only on the negative aspects of experiences of caregivers and HIV-infected men. Research participants commented that the focus on stressful events missed an important aspect of how they coped with the illness. To capture these experiences, participants were asked about positive meaningful events.

As noted above, *creating meaning* was an important dimension within the current study. Finding positive meaning in stressful situations has been previously reported, but it has received limited attention. Further work needs to be done to develop an understanding of the nature of events that are both positive and meaningful in the context of chronic illness, and to determine how those events help people cope with chronic stress. Previous research with children and families has also focused on the demanding aspects of caregiving (Jessop, Riessman, & Stein, 1988; Jessop & Stein, 1991; Ray & Ritchie, 1993). More recently, however, a number of

researchers, especially in the gerontological area, have criticized this focus on stressors and burden in caregiving (Cohen, Pushkar Gold, Shulman, & Zuccherro, 1994; Langer, 1993). There is growing recognition that caregiving holds potential satisfactions and benefits (Cohen et al.; Kane & Penrod, 1995; Nolan et al., 1996). Findings from the current study also reflect the positive side of caregiving. The concept of burden in caregiving will be examined in more detail later in this chapter.

Clearly, emerging conceptualizations of coping in the face of ongoing strain are supported by the findings of this current study. The recent move towards viewing chronic stress in the context of an individual's total life also holds promise for research in chronic illness. It points out that focusing attention on specific stressful events may neglect other important aspects of an individual's life that contribute to his or her ability to survive. However, our understanding of family coping in the context of chronic illness will remain underdeveloped if coping researchers continue to focus on individual coping efforts.

Family Coping

One of the underlying assumptions of family coping frameworks is that researchers can apprehend family adjustment and adaptation responses to stressful situations as a basis for developing interventions to reduce the anxiety and stress experienced by children and their families (Broome, 1998). As discussed under individual coping, however, life with a child who has an NLTI involves much more than responding to time-limited stressors that can be easily resolved. The Resiliency Model of Family Stress, Adjustment, and Adaptation (McCubbin & McCubbin, 1993) was discussed as part of the conceptual framework for this current study. It has now undergone further revisions and includes five levels of family appraisal. This revised model, the Resiliency Model of Family Adjustment and Adaptation (McCubbin, Thompson, Thompson, Elver, & McCubbin, 1998), continues, however, to focus on families during crisis situations.

Application of the Resiliency Model of Family Adjustment and Adaptation (McCubbin et al., 1998) to a family caring for a child with an NLTI would focus on family appraisal of their situation as a stressful event. For the families in the current study, established family patterns of functioning could have been assessed for adequacy in managing the adversity that occurs when a child is diagnosed with an NLTI. Assessment of family situational appraisals would probably have reflected the family's shared assessment of altered family routines and roles. As suggested by the model, many families apparently created new family paradigms to encompass changed roles and expectations and to reinforce the legitimacy of new patterns of functioning. An assessment of a family sense of coherence might have indicated the degree to which a family could change its existing or potential resources into useable resources that foster coping and adaptation. However, there is no established method of measuring family sense of coherence. Finally, assessment of a family's schema or world view might have assisted a family to find meaning by placing the situation within a broader set of values. However, the process of *navigating uncharted territory* encompassed much more than adaptation to crisis-induced stressors. For example, the extensive work involved in this process would have been missed if the focus was only on adaptation. It appears, therefore, that McCubbin et al.'s model is useful for examining family appraisal and coping responses associated with specific stressors, but has limited applicability in chronic illness.

Friedman and colleagues (Hanson, Kaakinen, & Friedman, 1998) noted that family health is most often conceptualized as family functioning or family adaptation. They suggested that no single theory or conceptual framework from nursing, family social science, or family therapy fully describes the relationships and dynamics of family life. Instead, family nursing requires an integrated approach to theory, research, practice, and education. The Friedman Family Assessment Model (FFAM) (Friedman, 1998) was designed to provide such an integrated model to guide family nursing practice. According to Friedman, Svavarsdottir, and McCubbin (1998),

wellness and functioning are optimized when adaptive responses and capacities are strengthened and encouraged, and actual or potential stressors are reduced. These authors acknowledged that family coping processes and strategies are essential if families are to adapt to the continual stresses and demands of family life. However, the basis of the FFAM (Friedman, 1998) is the recognition that understanding a family's experiences requires a holistic and comprehensive approach. Not only must one assess adaptational processes within families, but one must also acknowledge changes over time, growth, and disequilibrium, and recognize the importance of interaction between the family and its internal and external environments. In the opinion of the author of the current study, the theory of *navigating uncharted territory* could not have been developed without a similar appreciation of a holistic and comprehensive approach to understanding the experiences of families. This current study, therefore, provides support for the inherent value of Friedman's integrated approach to conceptualizing family functioning.

The Illness Trajectory

Coping frameworks are inherently reductionist in their perspective. Yet, life with a child who has an NLTi cannot be reduced to simply identifying the coping strategies used by individual family members, or even the family as a whole, when faced with acute or chronic stressors. Instead, the family's total biography and illness trajectory must be acknowledged. These families experienced an incredible workload because of the child's illness. They needed to manage the physical, cognitive, and emotional work that was generated by the situation, while continuing with daily life. Managing the work required a great deal of effort and extensive maintenance. Professionals, extended family, and friends seldom recognized the effort involved as families navigated this uncharted territory. The nature and shape of the illness trajectory uncovered in this process will be discussed in the following section.

The work of chronic illness has been well documented by Corbin and Strauss (1988). While this framework was developed only with adults, there are many areas that are relevant

when exploring the experiences of families whose child has an NLTl. This current study, therefore, extends the theoretical formulations proposed by Corbin and Strauss to include these families. The work of managing a chronic illness is described by Corbin and Strauss as a complex process and this was also true of families in this study. The work of chronic illness involves many different types of work, each accompanied by associated tasks. For example, care in hospital includes diagnostic work; technological work; comfort work; and clinical safety work. Associated tasks include controlling symptoms; monitoring, preventing, and managing crises; and managing limitations of activity (Corbin & Strauss, 1988). Illness work at home must also include biographical work, such as making the illness part of ongoing life; coming to terms with the illness, its limitations, and the possibility of death; and developing a new image of oneself, based on the illness and the associated bodily changes. Other types of work in the exhaustive list drawn up by Corbin and Strauss include occupational work; marital work; domestic work; work involved in handling social situations; and child care work. Information work is viewed as an extremely important form of work that is the basis for many of the other types of work. Another level of work that must be done is articulation work which is the organization and co-ordination of all of the other types of work.

What is significant about the findings of this study is the importance of understanding that the work involved for families as they cared for their children with NLTIs was not simply a list of tasks. This work, often with little support from others for an extended period of time, was carried out while families were grieving for the losses associated with the child's diagnosis, prognosis, and deterioration. Moreover, it is important to note that the demands of each task were not static. Families could not relax once they had learned one task, because, over time, the demand for a particular task changed or other tasks became necessary.

Corbin and Strauss (1988) identified three phases in the diagnostic period of chronic illness that were similar to the experiences of families in this current study. These researchers

conceptualized the three phases - prediagnosis, the announcement, and postdiagnosis - as the diagnostic quest. This diagnostic quest may be short or lengthy, and it involves work such as information work. It may also be repeated whenever there is a need to gather additional diagnostic information, such as if new symptoms appear. Uncertainty is a characteristic of the diagnostic quest. The quest often begins when the person acknowledges symptoms as important. Sometimes, symptoms are only recognized during a routine checkup or when receiving treatment for another disorder. In the prediagnostic phase, actions are taken to uncover the reason for changes. This phase varies depending on a number of factors including the nature of symptoms; when and how symptoms are reported; the skill of the investigating physician; the availability of technology; and the organizational structure. The delays, misdiagnoses, and inconclusive results of the diagnostic period of chronic illness noted by Corbin and Strauss parallel those of most families where a child has an NLTi. Family experiences related to learning about the child's diagnosis are also similar to those reported by Corbin and Strauss. In both instances, some physicians were sympathetic, while others were blunt. Reactions from patients, spouses, and families ranged from shock and disbelief to relief that they finally have a diagnosis. Most people needed time to absorb the meaning of the announcement. Finally, families used similar strategies in the third or postdiagnostic phase to find answers to questions that remain unanswered.

Clear parallels can be drawn between Corbin and Strauss' conceptualization (1988) of chronic illness and the experiences of families where a child has an NLTi. In addition, many of the identified types of work and their tasks were also present in the experiences of these families. It appears that Corbin and Strauss' framework may not only be applicable to adults with chronic illnesses and their partners, but may also be extended to families of children with NLTis. Unfortunately, it also appears that, despite the available research and literature, professionals continue to minimize or ignore the energy required by individuals and families to manage their chronic illness. Professionals may not be aware of strategies that families can use to mitigate

their work. If professionals are cognizant of both families' experiences and potentially helpful coping strategies, then they may be more willing or able to acknowledge the families' work. They can then work with families to identify appropriate interventions to alleviate the workload.

Central to Corbin and Strauss' work (1988) was the concept of illness trajectory. They adopted the term trajectory to encompass not only the illness course, but also the organizational work that was done and the impact on those who were involved with the work. The illness trajectory was viewed as a way of recognizing the active role that people take in changing the course of an illness. Moreover, it captured experiences that involved time, work, nonmedical features, and the interplay of workers who included patients, spouses, and health care providers.

According to Corbin and Strauss (1988), an illness trajectory is shaped by the interaction between the disease process, the individual's response to the disease, family biographies, and trajectory projections and schemes that are offered by health care professionals and family members. They propose that trajectories have a shape that is variable in form and duration. In addition, phases, their combinations, and subphases give shape to the trajectory. These phases include acute, comeback, stable, unstable, and downward. The phases reflect the kind and amount of work that is needed to manage the illness, and suggest what the potential impact might be. There are a variety of downward phases, but all usually involve combinations of comebacks, unstable periods, or stable plateaus. The uncertainty of these combinations cause problems for those passing through these aspects of an illness trajectory. Four illness trajectories that might result in death have been identified: certain death at a known time; certain death at an unknown time; uncertain death, but a known time when the certainty will be established; uncertain death and an unknown time when the certainty will be established. Families with a child who has an NLTi faced certain death at an unknown time as they navigated uncharted territory.

Experiences of navigating uncharted territory for families of children with NLTIs were not related to steady decline in the child's health. Instead, families lived their lives on plateaus of

relative stability, waiting for the next crisis or sudden decline in their child's condition. During each crisis, families dropped off the current plateau and fell until they reached the next plateau. Rolland (1994) has also noted that progressive illnesses are often characterized by a stepwise or progressive deterioration, and that progression may be rapid or slow. He suggested that patients and caregivers get minimal relief from symptoms during progressive illnesses as they have to constantly adapt and adjust to role changes. While this uneven and stair-like trajectory of certain death at an unknown time has not been previously documented in pediatric research, the idea of plateaus and drop offs is partially supported by the literature. Previous research with parents of chronically ill children has identified critical times in a child's illness when families face an increase in needs or when there are changes in the support structure (Clements et al., 1990; Whyte, 1992). These events precipitate periods of crisis and disequilibrium that are similar to those reported by families in this study. In addition, researchers concur with findings from this study that additional professional assistance is often required by families at those times (Clements et al.; Whyte).

Family researchers also support the concept of plateaus during illness experiences, though indirectly (Clements et al., 1990; Whyte, 1992). They describe families as living in a state of equilibrium when emotional and physical support is available to meet their needs. Other researchers have identified feelings of isolation and loneliness while living on the plateaus that are similar to those reported by families in the current study (Davies, 1996; Diehl et al., 1991; Walker et al., 1989; Woolley, 1991; Woolley et al., 1991). When families have little contact with health care providers in hospital, they often feel lost and alone. Many families have advocated for an ongoing relationship with one person who can co-ordinate care, provide guidance and develop a close and trusting relationship with them (Chambers et al., 1989; Davies; Diehl et al.; James & Johnson, 1997; Stein & Woolley, 1990; Woolley; Woolley et al.). Research on the effectiveness of such contact, however, is limited. In one study (Burke, Handley-Derry, Costello,

Kauffmann, & Dillon, 1997), researchers used a two-group, pretest-posttest design to determine the efficacy of a community-based, stress-point nursing intervention with families of chronically ill children. A research nurse worked with parents in the experimental group to identify their stresses and to develop coping strategies. She maintained contact with families by telephone, mail, and face-to-face meetings. Results showed improved coping and family functioning when these families received the intervention (Burke et al.).

There is little research that examines the trajectory of certain death at an unknown time. Copp (1996, cited in Copp, 1998) appears to be the only previous study that explores this trajectory. Apart from her study, the nature of the form, shape, and duration of this death trajectory has not been articulated in any depth. This current study adds to the available knowledge about the trajectory of certain death at an unknown time.

Gender, Communication, and Culture

Parents clearly identified many factors that influenced their ability to care for their children. However, they did not typically recognize the influence of gender on their behaviours and on their communication styles with partners. No previous research was found that explicitly explored gender differences in palliative care, though at least one researcher has recently suggested exploring gender differences in bereavement research (Stroebe, 1998). It is proposed that, in this current study, the differences between parents, their communication difficulties, and the impact this had on the illness trajectory can be explained, at least in part, by the complex interactions between gender, culture, and communication. Gender identities are created by communication. In addition, communication is used to express gendered identities in interactions with others. Moreover, gender reflects the culture and values of society (Arliss, 1991; Kalbfleisch & Cody, 1995; Wood, 1994).

Symbolic interactionism suggests that gendered identities are learned through interaction with one's own society (Mead, 1934). Particular behaviours are reinforced or discouraged to

ensure effective socialization into the culturally acceptable gender. For example, boys are often told to "keep a stiff upper lip" and not cry, whereas girls are encouraged to express their emotions. Communication is one of the primary ways in which societal structures are defined and maintained. Individuals can reinforce cultural prescriptions for gender by accepting the traditional views and behaving in expected ways. Conversely, individuals can challenge the accepted norms and attempt to change societal views. Thus, communication plays a vital role in both perpetuating existing gendered identities and in redefining them (Arliss, 1991; Kalbfleisch & Cody, 1995; Wood, 1994). In addition, society assigns roles to individuals and expects those roles to be filled. For example, women are still regarded as caretakers, whereas men are regarded as the primary breadwinners for families. Individuals internalize these roles and behave in ways that communicate their understanding of their gendered identities (Arliss; Kalbfleisch & Cody; Wood). Thus, mothers' and fathers' roles are socially constructed and should not be stereotyped. Further, illness trajectories are shaped by these social constructions and gendered identities.

In addition, some theorists (Arliss, 1991; Kalbfleisch & Cody, 1995; Wood, 1994) suggest that thin or fluid ego boundaries partially explain why women are often empathetic, sense others' feelings and experience them as their own, become so involved with others that their own needs are neglected, and feel responsible for others. Similarly, rigid boundaries might also explain why men keep distance between themselves and others, are less likely to experience others' feelings as their own, and rarely take responsibility for other people.

Given what is known about gendered identities, it is not surprising that gender differences were present in this study. Recent research with families of medically fragile children (Patterson et al., 1994) and children with developmental disabilities (Heaman, 1995) has also reported differences between mothers and fathers. However, these authors did not explicitly link the differences to gendered identities. In the current study, there were often differences in how mothers and fathers grieved, in how they responded to medication and feeding regimes, in the

amount of information they required, and in how well they cared for themselves. It seems that these differences reflect the respective gender identities and ego boundaries that have been previously identified. In addition, the assigned societal role of mothers as caretakers was fulfilled, even by mothers who did not want to stay at home.

It is important for health care professionals to recognize that gendered identities exist and to acknowledge the role they might play in parents' behaviours. Instead of perpetuating stereotypes based on sex, professionals need to be aware of the dimensions of gendered identities so they can provide the most appropriate interventions to families. Recent research supports the suggestion that parents do not always behave in stereotypical ways (King, King, & Rosenbaum, 1996; Nagy & Ungerer, 1990). If professionals are self-reflective and critical of their own expectations, they may recognize biases that influence their interactions with families. For example, health care providers may realize that they make some mothers feel guilty because they strongly encourage mothers to stay at home with their children. Or they may find that, because of their expectations about men, they have failed to recognize how deeply fathers are affected by a child's illness. It is also important for professionals to help parents understand the influence of gender identities on their experiences so parents can better understand each other's behaviours.

Gendered identities are also reflected in communication styles. In this study, parents often had difficulties in communicating. In many instances, they were upset because they did not understand why their partner's reaction to the situation was so different from their own. As also noted by Clarke-Steffen (1997), discrepant views were frequently a source of conflict. Some parents were able to accept differences and learned to better understand their partner. Diverse communication styles have been identified by a number of authors (Arliss, 1991; Kalbfleisch & Cody, 1995; Wood, 1994). Wood suggests that understanding and appreciating diverse communication styles is important because it allows improved interaction and communication.

Communication itself has two levels of meaning: the content or literal meaning, and the relationship level of meaning (Wood, 1994). The relationship level of meaning is complex and defines the relationship between communicators. It is important because it provides the context in which literal meanings are interpreted. Recognizing the relationship level of meaning is particularly crucial when examining patterns of communication across genders. Furthermore, human communication is symbolic. Thus, meanings are created and interpreted through abstract, and frequently ambiguous, symbols that are used to represent phenomena. Each partner in a communication creates and interprets the meaning of the communication through the lens of personal experience and values. Clearly, communication can become very confusing and difficult because of the possible multiple interpretations (Arliss, 1991; Kalbfleisch & Cody, 1995; Wood).

Language is a tool for communication, but language also constitutes cultural views of masculinity and femininity. It contributes to defining people as masculine or feminine and these identities are reflected in a person's style of communication. In general, language is used in different ways based on one's socialization into masculine and feminine speech communities (Arliss, 1991; Kalbfleisch & Cody, 1995; Wood, 1994). Masculine speech patterns tend to emphasize achievement and goal attainment. The purpose of communication is to show independence and strength. Feminine speech patterns reflect efforts to be collaborative and to develop relationships. Therefore, the general purpose of communication in this case is to establish and maintain relationships with others (Wood).

Generally, women like to talk because they view communication as fostering relationships and understanding. Women's speech patterns tend to emphasize equality between speakers, show support for others, use inclusive language, demonstrate responsiveness, and employ tentativeness as a strategy to keep conversations open (Wood, 1994). Men, however, generally tend to give advice, focus on achieving instrumental objectives, dominate the

conversation, use forceful and authoritative language, speak in the abstract rather than from personal experiences, and give few response cues (Arliss, 1991; Wood). Men often choose not to talk because disclosing personal information may be seen by other men as a sign of weakness. In addition, when men wish to support or comfort others, they tend to avoid talking. To men, talking shows disrespect for another's independence and suggests that the other person is weak. Clearly, when men and women have different communication patterns and different goals for communication, then the stage is set for misunderstanding and miscommunication.

Differences in communication styles between parents were found extensively throughout this study. They frequently led to conflict and misunderstanding between parents and posed a threat to the survival of the family. Some parents were unable to resolve these differences and the marital unit was severely strained. Parents who learned to understand that having differences was alright and who discovered satisfactory ways to communicate found that tensions were reduced and the family became more stable. As Wood (1994) suggested, understanding and appreciating diverse communication styles allowed improved interaction and communication. Health care providers who also understand different communication styles may be more able to communicate effectively with families. In addition, they may be able to support and guide families in their efforts to solve communication difficulties.

Family-Centred Care

The discussion in this next section centres around the concept of family-centred care and the influence of health care relationships. Despite the increasing conceptual acceptance of family-centred care, the practice of family-centred care has not always been optimal (Broome, 1998). Based on the findings from this study, challenges to family-centred care for families of children with NLTIs arise from both the current arrangement of the health care system and from some practitioners within that system. While all health care professionals now recognize that care of a child must also include the family, pediatric physicians have traditionally focused their

attention on the individual, rather than the family (Bowden, Dickey, & Greenberg, 1998). Family centred-care is a philosophy of care that has increasingly been adopted, especially in pediatric nursing. This approach to families arose from recognition that children cannot be seen as separate and distinct from their families. Various definitions of family-centred care emphasize that the focus of care should be the family (Bowden et al.).

For many people, the health care system is seen as insurance against death (Thorne, 1993). Until they become involved in it, they do not understand that a system which is set up to deal with acute, curable illness cannot provide adequate care for those with chronic or incurable diseases. Similar to previous studies (Davies, 1996; Diehl et al., 1991; Gibson, 1995), families in this current study suggested that the acute, curative system fails to address their needs. Moreover, the current system is fragmented and highly bureaucratic. For example, families needed to deal with various branches of government before they could obtain needed respite services. Yet, respite from the constant care of the ill child was crucial for families. As has previously been reported by many other researchers (Davies; Diehl et al.; Folden & Coffman, 1993; Gravelle, 1997; Sherman, 1995; Snowdon, Cameron, & Dunham, 1994; Stein & Woolley, 1990; Treneman, Corkery, Dowdney, & Hammond, 1997), respite allowed families time for pursuits other than caring for the ill child. Benefits of respite, such as time to catch up on sleep, reduced stress, and time for other activities, have been well documented in the literature (Davies; Gravelle; Stein & Woolley). Respite care is essential for promoting family health. Yet, families continue to report a shortage of available respite.

From a family-centred perspective, promotion of family health is seen as a major goal in provision of care. Conversely, when the focus is on the ill child, then care that is not provided directly to the child might be viewed as tangential and unimportant. Professionals and policy makers typically recognize some benefits of respite, but do not appreciate the extent to which respite is vital to the continuing functioning of families. While time to rest is important, the

opportunity that respite offers for communication between family members, especially parents, is largely unrecognized by professionals. Yet, parents report that finding the time to communicate is essential in ensuring continuance of their marriage and, subsequently, the family unit. Without opportunities to talk with one another, parents become estranged and families fall apart. Respite care, therefore, promotes family health in many dimensions.

The results of this study indicate that most families want to keep their child at home as much as possible, and they go to great lengths to do so. Yet, the health care system is focused on providing care in institutions. Moreover, despite an increasing contention from government that services should be delivered at home where possible, minimal support and funding is directed to community settings. Families often end up in institutions where even the physical layout makes it difficult for them to actively participate in care and decision making. Lack of space for family conferences can hinder the enactment of family-centred care. Many hospital rooms are small and not capable of holding multiple members of a family, so family visitors are restricted. Even when there is sufficient room for daytime visits, few hospitals are equipped for parents and other members of a child's family to stay overnight. Usually there are facilities for only one parent to spend the night. In such situations, it is difficult for the whole family to remain with the child. Further, professionals face difficulties in providing family-centred care when they have few opportunities to meet and work with the whole family. Families' experiences suggest the need for a critical analysis of the nature of health care in current society. In addition, the norms of health care professional behaviours typically reflect the system in which they occur. Therefore, the influence of the health care system on professionals and how that shapes families' experiences must also be examined.

Few health care professionals learn how to provide family-level care. Many are uncomfortable in discussing sensitive issues, such as sex, finances, terminal illness, or death. They shy away from talking about feelings and reactions. Yet, these and other psychosocial

issues are often critical areas for families. Life with a child who has an NLTI is emotional and poignant. The experiential nature of the chronic illness is very important to families. Yet, health care providers often minimize or disregard the meaning embedded in the experience. Other authors (Clarke-Steffen, 1993; Thorne, 1993) have also noted that individuals and families who are affected by chronic illness stress the importance of the experiential. Conversely, professionals focus on less abstract and more objective matters, such as providing analgesics to relieve pain. It may be that some professionals are uncomfortable in dealing with psychosocial issues, such as the meaning of illness, because they are unable to offer solutions. When they cannot provide answers or assistance, they may avoid the situation by focusing on an area where they can be successful. Yet, open and honest discussion of psychosocial issues was critical for most families. The only professional who typically provided an opportunity for such discussion was a psychologist. However, this crucial role was not supported by government funding in jurisdictions in which families in this study resided. Professional education and training, in conjunction with health policies, constrained and influenced families' experiences.

Health professionals frequently failed to acknowledge or understand the significance of the diagnostic process and event. Yet, all parents in the current study remembered vividly the manner in which the diagnosis was imparted. Some were still preoccupied with this event many years later. Parents valued an open, sympathetic, direct, and uninterrupted discussion of the diagnosis in private. Moreover, they wanted sufficient time for them to take the news in and for physicians to repeat and clarify information. Despite their intense dislike of evasive or unsympathetic brief interviews, many parents reported that the diagnostic process and event were controlled by professionals who avoided questions and who were insensitive to the impact of their discourse on families. These parents were not alone in this kind of experience. Other individuals and families have reported similar and equally unsatisfying encounters (Clarke-

Steffen, 1993; Knafl, Ayres, Gallo, Zoeller, & Breitmayer, 1995; Stein & Woolley, 1990; Thorne, 1993; Woolley, Stein, Forrest, & Baum, 1989).

Despite the tenets of family-centred care, power and responsibility for decision making often remains with the professionals. Families, therefore, are dependent to some extent on professionals. Their ongoing personal relationships with health care providers become important to families who are in longterm contact with the health care system (Baine, Rosenbaum, & King, 1995; Rosenbaum, King, & Cadman, 1992; Thorne, 1993). In this study, the calibre of relationships with health care providers significantly influenced families' experiences. Families who had open and honest relationships reported less conflict with professionals and more satisfaction with the care they received. In many cases, however, parents perceived professionals to be wary of parental involvement in the child's care and holding a dislike of co-operative models of health. This lack of acceptance of family participation concurs with previous findings that professionals often display a neutral or negative attitude towards collaborative care with parents (James & Johnson, 1997; Johnson & Lindschau, 1996). Parents of children with chronic disabilities have also described similar difficulties in relationships with health care professionals (Diehl et al., 1991; Walker et al., 1989). Despite the family-centred care philosophy espoused in many pediatric settings, clinical practice too often reflects the historical imbalance of power between health care professionals and those who require their services. One challenge for health care providers may be to recognize the important contribution of families and to shift the major responsibility for decision making from professionals to families.

A shift in responsibility to parents, however, also holds the potential for increasing parents' already heavy workload. There has been an increase in qualitative research during the past decade that has seen a change in the conceptual and organizing themes previously used in chronic illness. Deficit views have been replaced by analytic frameworks that are more optimistic and emphasize the competence of persons with chronic illness (Thorne & Paterson,

1998), or focus on the strengths of children and families in response to illness (Broome, 1998).

In a recent meta-study of qualitative, chronic illness research, Thorne and Paterson noted that this shift in perspective has resulted in an idealized picture of patients with a chronic illness. The patient is portrayed as competent, strong, and powerful, but the mundane aspects of chronic illness, such as pain and disability, are ignored. Thorne and Paterson suggest that, in an effort to reconceptualize clients from patients to partners, recent research fails to recognize the complexity of chronic illness. Moreover, this research minimizes or ignores the overwhelming work involved in chronic illness and does not acknowledge the burden that can be created by a partnership model of health care relationships. Professionals are also at risk of ignoring the probability that some patients continue to need expert advice.

The findings from this meta-study (Thorne & Paterson, 1998) reverberate in the current study. While parents in this study wanted to have some control of the situation, they were often overwhelmed with the exhausting and extensive work of illness. They expressed an immediate and urgent need for information. Parents spent considerable time and energy searching for information and learning about the illness. Most needed honest and appropriate information from health care providers to help them manage their situation. However, there is little information available about NLTIs. The scarcity of information may contribute to increased feelings of uncertainty and overload in these families. In addition, not all parents will have the internal and external resources to acquire the sophisticated knowledge that is required to be such a committed partner in health care. The time and effort involved in keeping current and taking the major responsibility for the child's care is not often recognized by professionals.

Decision making in health care relationships has been explored by a number of researchers (Beaver et al., 1996; Hack, Degner, & Dyck, 1994; Kirschbaum & Knafl, 1996). These studies consistently show that patients and families want varying amounts of control in decision making. It has been suggested that promoting involvement in decision making for those

patients who want involvement can lead to increased satisfaction with care (Davison & Degner, 1998). One intervention study showed that providing decisional support for patients with cancer was an intervention strategy that could be successfully incorporated even into a busy oncology clinic schedule (Neufeld, Degner, & Dick, 1993). This strategy was based on the concepts of commitment and control and used a simple technique for determining the degree of involvement the patient desired. The emphasis was on the patient's agenda, including assessing to what extent she wanted to participate, helping her to identify questions, and supporting her in obtaining the information she wanted and needed.

Some work has been done on measuring how much control a patient wants in health care decision making. The Control Preferences Scale (CPS) has been offered as a clinically relevant, easily administered, valid, and reliable measure of preferred roles in health-care decision-making (Degner, 1998; Degner, Sloan, & Venkatesh, 1997). However, knowing a patient's or family's preference for participation in decision making may not ensure that the appropriate level of participation will be offered by professionals. It is likely that a mismatch between the amount of participation desired by families and the level of participation offered by professionals would be associated with increased frustration and dissatisfaction with care. Therefore, Thorne and Paterson's (1998) suggestion that professionals learn not only when patients want to be treated as partners, but also learn when patients want to direct the actions of health care providers or give control to professionals merits further attention.

Trust and confidence in health care relationships were also extremely important for families in the current study. However, parents acknowledged that many professionals were unable to meet their requirements for trustworthiness. Competence and communication skills were necessary for trust, but were not sufficient in themselves. Professionals also needed to demonstrate respect for the families and recognize limitations in their own expertise. Similar results have been found by other researchers when families are dealing with chronic illness

(Clarke-Steffen, 1993; James & Johnson, 1997; Knafl, Breitmayer, Gallo, & Zoeller, 1992; Patterson et al., 1994; Thorne & Robinson, 1988b). One of the most valuable things a professional may be able to do is listen to parents and accept their concerns, because parents know their child better than anyone else (Clarke-Steffen, 1997; Cohen, 1995; Diehl et al., 1991).

Thorne (1993) noted that relationships with health care providers are inherently conflictual social processes. She reported that patients with chronic illnesses and their families are often dissatisfied with the attitudes of professionals. In addition, their encounters with professionals throughout the course of the chronic illness reframe their previously held views about health care providers. The process of health care relationships evolves over time and involves predictable shifts in emotions, attitudes, and behaviours (Thorne & Robinson, 1988a; Thorne, 1993). According to Thorne, most people with chronic illnesses initially believe that complete trust in professionals is necessary in order to get well. They are naïve in their assumptions that professionals will help them solve their problems. Inevitably, they lose their naïve trust and become disenchanted with the lack of assistance and support they receive from professionals. Some people are hostile and aggressive at this time, while others view it as a logical outcome of their early naivete and ignorance. Eventually, patients form a guarded alliance with some health care providers in order to obtain the health care required. The parents in this current study also described changes in their relationships with health care providers that reflected Thorne's process of naïve trust, disenchantment, and guarded alliance. It may be that this process of health care relationships is common to both chronic illnesses and progressive, life-threatening illnesses.

Impact of the Experience

The families' experiences had many impacts. Specific examples, such as physical, psychological, spiritual, or financial impacts, have been described in the previous chapter. In this

section, key themes that emerged from the study will be discussed including burden, chronic sorrow, fatigue, and family management style.

Burden

The immense amount of work involved when a child has an NLTi may appear overwhelming to someone outside of the family. One might quickly assume that families are severely burdened by their load. However, parents were very clear that caring for their children was not a burden. They were only doing what needed to be done for the children they loved. In fact, despite acknowledging the negative impacts on their lives, many parents identified satisfying aspects of the experience. In the past, research has focused on the demanding aspects of caregiving (Jessop et al., 1988; Jessop & Stein, 1991; Ray & Ritchie, 1993). However, a number of caregiving researchers, especially in the gerontological area, have recently criticized the predominant focus on stressors and burden (Cohen et al., 1994; Langer, 1993). A parallel may be drawn with the recent shift in research with people who have chronic illnesses. Thorne and Paterson (1998) noted that conceptual and analytic frameworks are moving from examining loss and burden towards a more positive focus that emphasizes the competence of persons with chronic illness. There has also been a growing recognition, as well as some empirical evidence (Cohen et al.; Kane & Penrod, 1995; Nolan et al., 1996), that caregiving holds potential satisfactions and benefits.

Nolan et al. (1996) suggested three reasons to explain the lack of research into sources and types of satisfactions experienced by family caregivers. First, stress and coping frameworks tend to pathologize caring, so caregivers are viewed as victims under stress. Second, family caregivers are sometimes seen as well-meaning, but potentially overprotective of relatives. Hence, services are offered with the intent of ensuring that the recipient of care can maintain some independence from the caregiver. Third, the historical tendency to view caregivers as passive recipients of services has led to the development of services that are based on

professional perspectives, rather than on caregivers' statements of what they actually do or need. Yet, uncovering the diverse sources of satisfaction experienced by caregivers would allow for a more complex and more complete understanding of the caregiving experience.

A number of themes have emerged from the literature about the potential satisfactions in caregiving (Nolan et al., 1996). Reciprocity and relationships are extremely important to satisfaction. Reciprocity does not imply the equal exchange of giving and receiving, but is embedded in relationships. Indeed, caring often grows out of established relationships. For reciprocity to be possible, the person receiving care must be seen as a valued person, rather than as a problem. In addition, there must be some meaning or worth attached to the caring. Given the typical parent-child relationship, it is not surprising, then, that parents of a child with an NLTI do not perceive caring for the child as a burden.

The importance of relational aspects of caregiving were evident throughout the current study. For example, most parents derived great satisfaction from the interpersonal dynamic between themselves and their child. Their pleasure over seemingly insignificant events, such as raising a smile from the ill child, supports Nolan et al.'s assertion (1996) that seeing the cared-for person content is a source of satisfaction for caregivers. Caring for someone else out of love, not duty, is another source of satisfaction that was voiced by parents in this study. According to Nolan et al., expressions of appreciation are also valued by caregivers. While parents often could not receive signs of appreciation from the child, they highly valued appreciation from others such as extended family, friends, or professionals. These votes of appreciation allowed families to continue with their caregiving roles.

Some other sources of satisfaction for caregivers arise from their ability to promote positive or avoid negative consequences for the cared-for person (Nolan et al., 1996). Many caregivers promote positive consequences by providing the best care possible. They often have a strongly held belief that they can provide better care than anyone or anywhere else because they

know the person so well. This attitude was very evident in the families in the current study. They were extremely proud of the care they provided, and sometimes credited the care with keeping their child alive. Moreover, they were able to delineate the kind of care that met their standards. Yet, caregivers who believe that no one else can provide adequate care may be reluctant to accept outside help. These caregivers, therefore, might continue caregiving despite the costs. Parents in this study suggested though, and this is supported by Nolan et al., that alternative services seldom provide care that meets caregivers' acceptable standards. One could argue that caregivers' standards are unrealistically high. Yet, their standards are based on meeting the needs of the people for whom they are caring. Surely, that should also be the basis for alternative services? Reassessment of current services and creation of acceptable alternatives must receive a high priority from practitioners and policy makers.

The perspective on burden that emerged in this study reflected more recent positions that providing care to a person one loves is not an onerous, thankless job. Rather than feeling compelled to remain in a caregiving situation that was not of their choosing, these families voiced a strong conviction that they chose to continue providing care because they loved their child. Further, many parents identified satisfying aspects of the experience. The findings in this study concur with other research and suggest that further research is required to identify the sources and types of satisfactions in order to obtain a more complete understanding of the caregiving experience.

Chronic Sorrow

Whether on or dropping off plateaus, parents in this study experienced a constant, ongoing grief that intensified or diminished in response to the current situation, but that never went away completely. While continuing grief is often associated with other chronic illnesses (Gibson, 1995; Gravelle, 1997; Parker, 1996), most researchers emphasize that feelings of grief arise at diagnosis, disappear after a period of time, and then recur at critical times (Bluebond-

Langner, 1996; Clarke-Steffen, 1997; Thorne, 1993; Whyte, 1992). Families of children with NLTIs, however, face chronic and recurrent grief because of the many losses associated with the child's illness over the relatively short period of the child's lifetime.

The concept of chronic sorrow has been proposed to explain families' responses to chronic illness (Fraley, 1990; Phillips, 1991; Teel, 1991; Warda, 1992). This concept was first articulated in 1962 by Olshansky to describe the psychological reaction of parents of severely, mentally retarded children (Olshansky, 1962). The recent development of a middle-range nursing theory of chronic sorrow is useful in examining families' grief responses when a child has an NLTi (Eakes, Burke, & Hainsworth, 1998). Chronic sorrow is defined by Eakes et al. as the "periodic recurrence of permanent, pervasive sadness or other grief-related feelings associated with ongoing disparity resulting from a loss experience" (p.180). According to these researchers, chronic sorrow is a normal response to ongoing disparity due to loss. The primary antecedent of chronic sorrow is an experience of significant loss. The second major antecedent is an unresolved disparity that is created when an individual's current reality is very different from the idealized reality. Trigger events emphasize disparities and force individuals to confront the gap between reality and expectations. For families in the current study, the initial loss came when the child was diagnosed with a life-threatening illness. Times of crises, such as when the child was in danger of dying, or significant milestones, such as birthdays, were clearly trigger events that caused increased grief and feelings of sadness for parents. However, families in this study were continually faced with losses and disparities. As one mother noted, parents often had little time to adjust to one loss before they were faced with another loss. In addition, families were surrounded by signs that life was not normal, such as equipment required by the child. Therefore, it is not surprising, that, unlike other families facing chronic illness, many parents of children with NLTIs had an episode of grieving virtually on a daily basis. While the concept of chronic sorrow provides a framework for understanding these parents' responses, the almost daily losses

experienced by these families are not typical of all individuals facing ongoing disparity and loss. The extent of loss and disparity and the resulting grief has seldom been recognized by health care providers.

Fatigue

Most parents in this study were exhausted. They were on call 24 hours a day, 7 days a week. Many parents reported that they suffered through months and years of not sleeping. Parental fatigue has been reported in other studies where a child has a life-threatening illness (Gravelle, 1997; Martinson, 1993; Stein et al., 1989; Stein & Woolley, 1990). Little attention has been paid, however, to the impact that fatigue has on these family members and the family as a unit. Fatigue is increasingly prevalent in today's busy society (Loge, Ekeberg, & Kaasa, 1998; Walters & Denton, 1997). It is also an extremely common symptom reported by people with a variety of physical and psychological diseases (Breitbart, McDonald, Rosenfeld, Monkman, & Passik, 1998; Bruera & MacDonald, 1988; Deatrick, Brennan, & Cameron, 1998; Ford, Trigwell, & Johnson, 1998; Grady, Anderson, & Chase, 1998; Grohar-Murray, Becker, Reilly, & Ricci, 1998; Hockenberry-Eaton et al., 1998; Nail, Jones, Greene, Schipper, & Jensen, 1991; Pollina, Kaufman, Masur, & Krupp, 1998; van Servellen, Sarna, & Jablonski, 1998). Moreover, fatigue is frequently found in family caregivers (Herth, 1993; Jensen & Given, 1991; Lindgren, 1990; Nolan et al., 1996). Yet, it is only recently that researchers and practitioners have shown increased interest in understanding this phenomenon. There is a growing literature on the incidence and impact of fatigue when someone is ill. Similar research with family caregivers remains lacking.

One of the difficulties with research on fatigue is the confusion that continues to surround the concept (Ream & Richardson, 1996). Fatigue is used in diverse applications and may be both a noun and a verb. Its definition is unclear and it is frequently used interchangeably with other nouns, such as weakness or tiredness, or verbs, such as to exhaust or tire. No universal definition

of fatigue exists (Winningham et al., 1994), though characteristics of fatigue include that it follows physical exertion or sleep deprivation, is associated with physical or mental exhaustion, causes decreased functional ability, and often involves feelings of discomfort (Ream & Richardson). Health care definitions of fatigue usually add that fatigue is a multicausal, multidimensional, subjective phenomenon that affects the whole person (Piper et al., 1998; Winningham et al.). Few studies, however, have attempted to measure fatigue as a multidimensional sensation (Irvine, Vincent, Bubela, Thompson, & Graydon, 1991; Piper et al.).

Sleep deprivation and fatigue were prevalent in parents in the current study. The impact of fatigue included physical and mental exhaustion that manifested itself in a number of ways. The effects were often subtle and not explicitly recognized as being attributable to fatigue. Potential effects of fatigue included physical weariness, impaired cognitive functioning, slowed speed of information processing, and depression. Similar issues have been previously identified in both healthy and ill populations. Frustration, reduced performance as reflected in difficulties completing complex tasks, and increased error rates have been noted when fatigue was present in healthy pilots (Chelette, Albery, Esken, & Tripp, 1998), air traffic controllers (Luna, 1997; Luna, French, & Mitcha, 1997), and transportation workers such as truck and train drivers (Ellingstad, 1998). Ellingstad warned of the adverse effects on transportation safety and the potential dangers arising from operator fatigue. Moreover, he stated that the far-reaching and debilitating effects of fatigue are serious and continuing, yet most people fail to understand the problems associated with fatigue. In families of children with NLTIs, these effects could result in actions such as parents administering incorrect medications or failing to follow correct procedures when using medical equipment. Clearly, the potential for serious negative consequences increases the longer parents suffer from sleep deprivation and fatigue.

Fatigue has also been associated with affective, cognitive, and behavioural responses that include irritability, impaired cognitive functioning, and inability to concentrate on daily tasks

(Ream & Richardson, 1996). These responses were evident in families in the current study. One particular aspect of cognitive functioning, attentional impairment, was especially striking. This aspect was exemplified in the strategy of *going into slow motion*. Some families reported that they slowed down their pace and went into slow motion so that they could manage their lives. However, *going into slow motion* also affected their ability to learn new things or to process information.

Attentional theory provides a framework for understanding this attentional impairment (Kaplan & Kaplan, 1982). Attention is the ability to focus and concentrate or to direct attention. Kaplan and Kaplan proposed that directed attention requires mental effort. In addition, one requires a minimal quantity of attentional resources in order to process information. When resources are reduced, then attentional fatigue results and information processing slows down. Fatigue itself has been identified as one cause of reduced resources (Matthews, Davies, & Lees, 1990). In addition, multiple factors may increase demands for directed attention over long periods of time. When demands exceed available resources, then a person may be at risk for attentional fatigue (Cimprich, 1992a; Cimprich, 1992b; Cimprich, 1995; Jansen & Cimprich, 1994). Family of children with NLTIs faced both fatigue and increased demands on their attention. The fatigue may have reduced their attentional resources. Moreover, they provided highly vigilant care and performed multiple, complex tasks that could have reduced their effectiveness in basic cognitive activities, such as acquiring information. The impact of *going into slow motion* has implications for professionals. As noted by the parents themselves, families in slow motion may need extra time to work through new ideas or suggestions until they finally understand them and no longer find them stressful. It then becomes important for professionals to slow down as well to match the slower pace of families. The concept of *going into slow motion* has not received attention in the literature. However, in a study of terminally ill adults receiving palliative care in a hospital setting, patients also slowed down their pace in order to

make sense of their situation. Nurses who were more effective at providing care slowed down their pace to match that of the patients (B. Davies et al., 1998). Further exploration of this concept and the applicability of attentional theory may provide added insights into the experiences of families when a child has an NLTI.

A number of authors have demonstrated a relationship between fatigue and depression (Breitbart et al., 1998; Ferrell, Grant, Funk, Otis-Green, & Garcia, 1998; Irvine, Vincent, Graydon, & Bubela, 1998; Messias, Yeager, Dibble, & Dodd, 1997; Piper et al., 1989; Rose, Pugh, Lears, & Gordon, 1998). Fatigue is a well-documented symptom of depression. Although some authors have conceptualized that depression can be secondary to fatigue in some cases, this relationship has not been clearly established (Schneider, 1998). Some evidence does suggest, however, that fatigue can lead to depressed mood. A couple of parents in the current study perceived that fatigue induced feelings of depression. These relationships are further complicated by the possibility that attentional performance may also be influenced by the diminished arousal associated with depression (Cimprich, 1993). There is a need to clarify the complex relationships between fatigue, depression, and attentional capacity in order to more fully understand their effects and to determine appropriate interventions.

In clinical populations, fatigue has been shown to adversely affect one's quality of life (Breitbart et al., 1998; Ferrell et al., 1998). In addition, it has been correlated with increased illness uncertainty (Mast, 1998) and reduced feelings of healthiness or well-being (Leddy, 1997; Messias et al., 1997). The development and evaluation of clinical interventions to relieve fatigue and improve quality of life is imperative. If depression can be caused by fatigue, then relief of fatigue might also improve depression. While most of the existing fatigue research has been conducted with non-caregivers, it may offer some guidance for relieving the fatigue experienced by family caregivers. Research has shown that performance declines and errors increase during night-shifts (Luna et al., 1997). Respite care at night might prevent fatigued parents from making

mistakes. It might also allow parents to sleep and reduce their fatigue (Grohar-Murray et al., 1998).

Rest and recovery of directed attention may need to be encouraged (Kaplan & Kaplan, 1982). Voluntary or direct attention requires effort. Families require ways to reduce the need for directed attention. Involuntary attention that does not require sustained effort should be promoted. Professionals could encourage families to participate in activities that they enjoy and that will take their minds off the situation. Researchers have reported the successful use of distracting or diversional activities to relieve fatigue (Grohar-Murray et al., 1998; Nail et al., 1991). Simple restorative activities that have been suggested include walking or sitting in a natural environment, gardening, bird watching, or doing crafts or hobbies (Cimprich, 1993). Respite care may also allow family members time away from directed attention. Parents need not leave the house to benefit from respite care. When they are relieved of their constant vigilance, their need for directed attention may be reduced and they can then rest.

Physical exercise may be another way of relieving fatigue. Exercise has been used successfully by patients with diseases such as myasthenia gravis (Grohar-Murray et al., 1998) and cancer (Mock et al., 1994; Schwartz, 1998a) to reduce their fatigue, and to decrease their perception of fatigue (Mock et al.; Winningham, 1991; Winningham, 1992). A few parents in the current study, especially fathers, noted the benefits they received from exercise. Mothers were less likely to take the time to exercise. While promoting exercise may seem contrary to a desire to conserve energy in people who are already fatigued, exercise may actually reduce fatigue. Discussion about the potential benefits of exercise, plus provision of respite, may encourage families to increase their exercise and reduce their fatigue. Finally, cognitive-behavioural interventions have been used with patients to improve outcomes in rheumatoid arthritis (Sinclair, Wallston, Dwyer, Blackburn, & Fuchs, 1998) and chronic fatigue syndrome (Deale, Chalder, &

Wessely, 1998). Similar interventions might be useful to change families' perceptions and beliefs about exercise, activity, and fatigue and may result in improved outcomes.

Family Management Style

Although there was no intent to use the Family Management Style (FMS) framework in this current study, the parallels between the process of *navigating uncharted territory* and the management behaviours in the FMS could not be ignored. The FMS is a conceptual model that was developed from clinical observations, a concept analysis (Knafl & Deatrick, 1990), a review of the literature (Deatrick & Knafl, 1990), and a research project (Knafl, Breitmayer, Gallo, & Zoeller, 1987). Subsequent research has provided support for and further explicated this model (Clarke-Steffen, 1997; Knafl, Breitmayer, Gallo, & Zoeller, 1996). The emphasis of the model is on the reciprocal nature of defining and managing varying aspects of a family's response to chronic illness, as well as how those aspects are shaped by different members of a family. Management is defined as the "active, behavioral component of the family's response" (Knafl & Deatrick, p.8). The management behaviours chosen by individuals and families are influenced by their perceptions and definitions of the chronic illness. Management behaviours can be described in terms of their goal, that is the result toward which a family's effort is directed; the underlying conceptual dimension that defines the descriptive qualities or organizing framework that characterizes the behaviours of interest; the implementor, that is the person who orchestrates or carries out the behaviours; and the foci towards which the behaviours are directed. These foci include the child/illness, the family, and the social system. The current study both supports and extends the FMS framework. Support was found within the process of *navigating uncharted territory* itself. For example, *creating meaning* allowed families to select particular management behaviours. This study extends the FMS by describing another style of management. The families' choice of family management style reflected the intense impact that their child's illness had on their lives. Unlike most families whose children have chronic illnesses (Deatrick &

Knafl), these families were unable to use the FMS of normalization to manage their lives.

Instead, they needed to develop other strategies that allowed them to continue living with a child with an NLTI.

For most families with ill children, their goal is to have a normal life (Bluebond-Langner, 1996; Clarke-Steffen, 1997; Deatrick, Knafl, & Walsh, 1988). Families work hard at maintaining a normal lifestyle. They develop normalization strategies that allow them to appear like other families (Bluebond-Langner). For example, some families redefine what they perceive as normal to encompass the tasks involved in providing care to their children. Or they emphasize the areas in which their children are like other children, such as a child's crucial role on her soccer team. Conversely, families in this current study reported that their lives were not normal. They could not participate in normal activities. They were unable to function as they had done before their children became ill or as other families continued to function. Unlike families of chronically ill children who were relatively healthy (Bluebond-Langner), families in this study were unable to treat the child and siblings equally. Instead, parents focused heavily on the ill child and gave less than optimal attention to siblings. In addition, other relationships were put on hold or discarded because parents focused on the child. Families were also surrounded with constant reminders of their child's disease, such as oxygen and other equipment, because the child required complex care.

Some researchers (Bluebond-Langner, 1996; Deatrick & Knafl, 1998; James & Johnson, 1997) have identified a phase in non-neurological, life-threatening illnesses when a similar pattern appears and normalization strategies are no longer used. This phase is invariably near the end of the child's life. In most cases, the child has been relatively healthy and normal following diagnosis and treatment. However, at some point, the development of complications and an increase in complexity of care impinges on family life. As deterioration increases and care becomes more complex, there is little left that can be perceived as normal. During this phase,

families acknowledge that the child is going to die. They also discontinue the use of normalization strategies to meet the challenges of daily living (Bluebond-Langner; Deatrick & Knafl).

Families whose children have an NLTi, such as the families in this and other studies (Davies, 1996; Parker, 1996) move through a somewhat different process than other families where a child has a life-threatening illness such as cancer (Clarke-Steffen, 1997; James & Johnson, 1997), cystic fibrosis (Bluebond-Langner, 1996; Whyte, 1992), or other diseases (Gravelle, 1997). This difference may be related to the treatment available and the hope for a cure that families can reasonably expect. When a child has an NLTi, families may have less hope for a cure because of the nature of the disease. The neurological system is extremely complex, and cures for damaged brains have been very limited. Furthermore, the lack of research and funding for research limit the possibility of a cure being found in the near future.

In addition, families of children with NLTis must not only accept a life-threatening prognosis, but they must also accept the child's disability. In many ways, their lives are more similar to families of children with chronic disabilities than they are to families of children with other life-threatening illnesses. Many children who are ill are not also considered disabled. For example, a child with cancer is labelled as sick, but is not considered disabled. Children with NLTis, however, are disabled as well as terminally ill. They seldom have a period of relative health and normality once their illness manifests itself. Instead, they quickly develop disabilities that are usually visible and that affect family life. In addition, families are unable to escape or avoid the child's illness because of the amount of care and equipment the child requires.

Children with non-neurological illnesses and their families can usually manage their situations through accommodation and normalization strategies (Bluebond-Langner, 1996; Clarke-Steffen, 1997; Deatrick et al., 1988; Whyte, 1992). Yet, the opposite was true for families of children with NLTis. Those families who both accepted the child's terminal prognosis and

picked up the badge of disability were more able to manage the situation, whereas parents who wanted a normal life were less able to let go of the past and deal with the present. Whyte also suggested that families need to move through the transition from seeing themselves as a normal family to accepting that their family has a health problem. Normalization was a strategy that clearly served little useful purpose for families of children with NLTIs. It is not surprising, therefore, that these families developed other strategies to manage the work of living with a child with an NLTI.

There are interesting similarities between families whose children have NLTIs and families where an adult member is terminally ill. Adult families describe their experience as the transition of fading away (B. Davies et al., 1995; B. Davies, Reimer, & Martens, 1990). This transition starts with the ending when families realize that the patient is dying. During the ending, families redefine the patients as dying and no longer view life as "normal." This redefinition is necessary so that families can accept the patient's imminent death. A similar task is necessary for families of children with NLTIs, though they must redefine their child as dying and disabled. The transition continues for adult families as they enter the neutral zone. The neutral zone is characterized by fear, loneliness, confusion, and uncertainty. It is an uncomfortable place that families want to leave as quickly as possible. The neutral zone compares with the ongoing lives of families who are *navigating uncharted territory*. However, unlike adults in palliative care, these children usually have a life expectancy of more than six months. Therefore, families of children with NLTIs are stuck in this difficult and unpleasant zone for extended periods of time. These families also face some tasks that are similar to the adult families. They struggle with the paradox of living and dying at the same time; there are many changes to contend with; and they search for meaning in the experience (B. Davies et al.). Finally, some families live day to day and make the most of the time left. These families are

somewhat similar to the adult families. However, they are not yet preparing for the child's death. The beginning of the transition may only happen for these families when death is imminent.

The FMS framework was not part of the conceptual framework at the outset of this study. However, there are significant parallels between the FMS and the model developed in the current study. Further, the current study extended the FMS framework by explicating a management style that has not been previously described in detail elsewhere.

Summary

This chapter placed the findings of this newly developed grounded theory within the broader context of current research and literature. It was determined that existing stress, coping, and adaptation frameworks provide some direction for education, research, and practice. These frameworks, however, restrict one's view of families' experiences to only those aspects that involve coping and adaptation. In addition, most researchers have examined acute stressors, so little is known about coping with chronic stress. Managing a life-threatening illness was portrayed as a complex process in which the many different types of work and associated tasks are seldom recognized by professionals. The concept of illness trajectory encompasses the illness course, the organizational work, and the impact on those involved with the work. Trajectories have a variable shape due to form, duration, and phases. Families with a child who has an NLTI face a illness trajectory of certain death at an unknown time. There is little previous research that examines such a trajectory. The uneven and stair-like shape of this trajectory found in the current study has not been previously documented in pediatric research. It was proposed that the illness trajectory is also shaped by the interaction between gender, communication, and culture. Differences between parents, their communication difficulties, and the resultant impact on the illness trajectory were discussed and explained in relation to these complex interactions.

It was argued in this chapter that family-centred care has been increasingly accepted at the conceptual level, but the practice of family-centred care is not always optimal. Care is

frequently provided at an individual rather than family level, often because few health care professionals learn how to provide family-level care. It was suggested that the current health care system makes it difficult for families to actively participate in care and decision making. Further, some professionals are reluctant to work in collaborative partnerships with families. Conversely, some professionals may shift total responsibility to parents and not acknowledge the burden that can be created by a partnership model of health care relationships. The calibre of ongoing relationships with health care providers they trust and in whom they have confidence was reported as significantly influencing families' experiences. It was suggested that the process of health care relationships evolves over time and involves predictable shifts in emotions, attitudes, and behaviours.

Key themes that emerged from an exploration of the toll that the whole experience took on families included burden, chronic sorrow, fatigue, and family management style. It was suggested that caregiving does not need to be a burden. Further, the satisfying aspects of providing care to a loved one have been largely neglected in research because of the predominant focus on stressors and burden. The concept of chronic sorrow was expanded beyond its common emphasis on feelings of grief arising at diagnosis, disappearing after a period of time, and then recurring at critical times. Families with a child who has an NLTII experience ongoing loss, disparity and resulting grief that is seldom recognized by health care providers. Most parents were exhausted. It was reported that little attention has previously been paid to the impact of fatigue on family members and the family as a unit. Implications for professionals relating to fatigue in families were discussed. Finally, it was proposed that the current study both supports and extends the Family Management Style framework. Support is found within the process of *navigating uncharted territory* itself. The study also extends the framework by describing another style of management.

The background and conceptual framework for this study were laid out in previous chapters. A review of related literature was followed by an explanation of the research design and procedures. The findings of the study were described in Chapter V as the process of *navigating uncharted territory*. These findings were then placed into the larger context of research and literature in the current chapter. The research study will be summarized in Chapter VII. Implications for practice, education, and research will also be offered in this final chapter.

Chapter VII: Summary and Implications

Summary

Children with neurodegenerative, life-threatening illnesses (NLTI) account for a significant proportion of children requiring palliative care. Most of their care is provided at home by their families. Yet, there is a paucity of research that examines families' experiences when a child is dying at home. In this study, the procedures of grounded theory were used to generate a detailed, contextually grounded account of families' experiences of living with a child who has an NLTI, and how those experiences changed over time; the impact on families; and families' perceptions of factors that influenced their ability to care for their children. Data collection was facilitated through a pediatric hospice care program and a children's hospital after ethical clearance had been obtained from each facility and from the researcher's university.

The main method of data collection was in-depth interviews supplemented by participant observation in families' homes. Data collection ceased when theoretical saturation was reached. Each family member was interviewed individually, then the family was interviewed as a group. None of the ill children could be interviewed because of their limited ability to communicate. Initial data collection occurred over a period of one year. In the second year, the evolving analysis was shared with families and further data were obtained from several parents. Twenty-nine participants from eight families were interviewed or observed. Two families had two ill children. Six of the ill children were boys and ages ranged from 3-13 years. All the ill children had NLTI that would eventually lead to their death. Individual diagnoses were not identified in the study to protect the children's anonymity. The different illnesses were manifested in many ways, but common attributes such as changes in verbal ability, changes in motor skills, and the proliferation of feeding disorders meant that these families experienced similar opportunities and challenges as they lived with the illness.

Data analysis was concurrent with data collection. Management and analysis of transcribed field notes and interviews were facilitated by the use of a word processor computer program. The researcher used memos and diagrams to document the process and to capture relationships between categories and subcategories. Constant comparative analysis of each transcript and field notes was used to check for commonalties and differences both within and across data sources. The process involved in families' experiences was identified by linking action/ interactional sequences. In the first level of data analysis, "open coding," data were examined line by line to identify codes or words that captured the meaning of the events. Similar phenomena were given the same conceptual name and these concepts were then grouped into preliminary categories. Connections between a category and its subcategories were made during "axial coding" when the researcher asked questions of the data and compared concepts. Evidence of differences and variation added density and variation to the theory. A coding paradigm was used to specify a category in terms of the conditions that gave rise to it: context, intervening conditions, action/interactional strategies, and resultant consequences. Finally, the core category was selected through the process of "selective coding." The researcher systematically related this central category to other categories, validated those relationships, and filled in the categories that needed further refinement and development. The core category was named with a high level of abstraction, and the chosen conceptual label fit the story it represented.

Families moved through a process of *navigating uncharted territory* as they lived with their dying child. This process occurred within a broader sociocultural context that influenced families' lives. Professional care was provided within an *acute, curative health care system* that often failed to meet the needs of families where a child had an NLTII. Interventions typically focused only on the child, despite the families' view that family-centred care was critical. The system structures were designed to provide emergency and technological services, and to discover new curative procedures. Yet, these children could not be cured. Parents often became

frustrated by a lack of support or assistance. They felt adrift and alone when they had no individual health care provider as a primary resource. Parents spent precious time and energy searching for information and resources because professionals seldom offered guidance. They also faced inequities and territorial issues within the system. In addition, the *sociocultural environment*, as reflected in the behaviours and attitudes of health care providers, extended family, friends, and strangers, influenced and constrained a family's ability to act in certain ways.

The basic social process of *navigating uncharted territory* was initiated by a precipitating event, such as one parent recognizing a deterioration in the child's motor skills. Families were forced to begin a journey that was unlike the "normal" life journey. Parents tried to describe an experience that was unique, filled with constant poignancy, and indescribable. The experience was incomparable with other life experiences, no matter how difficult those other experiences may have been. These families looked for ways to deal with the unfamiliarity, uncertainty, and unpredictability of their lives. Families were travelling without the benefit of a "map" based on the experiences of many other families. Because relatively few families had gone before them, or at least had documented their experiences, these families were travelling in uncharted territory. While all families underwent the process of *navigating uncharted territory*, some parents also underwent a transformation that resulted in personal growth. Those parents who made peace with their emotions, accepted the situation, and created positive meanings out of the experience felt more in control of their lives, were not overwhelmed by emotions such as anxiety and depression, and believed that they had gained from the experience.

Emotions of fear, uncertainty, and grief gave impetus to the process. While these emotions were always present, they changed in intensity over time. Fear and uncertainty were stronger at the beginning when parents were aware that something was wrong with the child. In many cases, the fear lessened immediately after receiving a diagnosis, while the uncertainty remained stable or intensified. The first six to twelve months following the child's diagnosis were

ones of intense grieving for all families. Gradually, the intensity lessened, but the grieving never went away. Parents experienced many losses that contributed to their grief. They had to adjust to each loss. Yet, there was seldom time to adjust to one loss before another loss occurred. The magnitude of the distress caused by what might seem, on the surface, small or insignificant losses was not apparent to all professionals. While emotions gradually declined over time, they increased with subsequent precipitating events. Families lived much of their lives on plateaus of relative stability, but, inevitably, periods of instability originated in subsequent precipitating events in the process that led to families *dropping off the plateau*.

Living with a dying child had profound physical, emotional, and financial impacts on families and required them to undertake extensive physical, cognitive, and emotional work. The process of *navigating uncharted territory* was characterized by four dimensions - *entering unfamiliar territory, shifting priorities, creating meaning, and holding the fort*. These dimensions were overlapping and iterative. Parents used strategies within the dimensions to manage the associated work. The behaviours of families were also related to the stage of the child's illness. Changes in parental strategies were closely linked to pivotal events in the illness trajectory.

The dimensions and strategies that characterized the process were constrained and facilitated by four intervening conditions - *relationships with health care providers, availability of information, gender differences, and communication between parents*. Relationships with health care providers were very important to families. They were viewed as dynamic and evolving over time. Relationships that were unsatisfactory to families became strained and negatively influenced their ability to manage their child's illness. Yet, health care providers were often insensitive to the impact of their discourse on families. Parents also emphasized the importance of ongoing relationships with a few key individuals.

Information was helpful as families struggled to live with their child's illness. At first, they tried to understand what was happening. Then, they learned as much as possible so they

could provide the best care for their child. Information also helped to reduce their fears and uncertainties. It gave them some control over the situation. In most cases, however, little or no information was available about the illness because NLTIs are uncommon. Some parents initially suspected that professionals were withholding information. They realized over time, though, that there was a paucity of documented information about NLTIs. The lack of information was often frustrating to parents and increased their fears and uncertainties.

Gender differences sometimes led to conflict and misunderstanding between parents and posed a threat to the family's survival. The marital unit was severely strained when parents could not resolve differences. Tensions were reduced and the family became more stable when parents accepted differences and understood that it was alright to have them. The arguments caused by differences were often ongoing and of long duration, and were a major issue in some families. There were often differences in how mothers and fathers grieved; in how they responded to medication and feeding regimes; in the amount of information they required; and in how well they cared for themselves.

Communication between parents was the final intervening condition. Understanding one's partner's point of view was essential in ensuring the successful communication that was central in making marriages work. Discrepant views were frequently a source of conflict. When parents accepted differences, then discrepant views could also be a source of strength if the different perspectives were viewed as complementing one another. Successful communication, however, was often only achieved after months of arguments or misunderstandings. Existing communication skills were usually inadequate for the current situation. Parents often needed external assistance to help improve communication. They also used multiple strategies to enhance both opportunities to talk and communication skills.

This grounded theory captures the experiences of families where a child has an NLTI in a way that extends our knowledge. The findings reported in this study also have important

implications for health care practice, education, and research. Some of these implications will be discussed in the final section of this research report.

Implications for Practice, Education, and Research

Practice and Education

The creation of a health care system that is responsive to the needs of patients and families seems paramount. Complaints about the fragmentation, inaccessibility, authoritarianism, and impersonal nature of the current system deserve consideration. Many others have suggested that health care should have a greater impact in human terms (Danielson et al., 1993; Thorne, 1993). Improvements in the delivery of care that recognize the effect of illness on the total person, rather than only at the cellular level, might be a step towards increasing the impact in human terms. Such changes must include integration of the family into the current health care system. This integration will require time and effort, but change appears necessary.

The health care system may only become responsive to families' needs if there is a shift in focus. Health care providers might consider how to move their attention beyond instrumental care in order to recognize the full implications of an illness trajectory. For example, families caring for a child with an NLTI needed to feel appreciated. Telling families that they are doing a great job is something that is easy to do and costs no money. Yet, it has significant meaning for families and helps them to continue caregiving. Professionals might also enrich caregiving by working with families to identify and actively encourage those activities that caregivers currently participate in and view as important. Initiating discussion about meaningful events in the caregiving process may allow families to see the positives in their experience. In addition, professionals might identify new routines or activities and make suggestions to families, based on their knowledge of what the particular family views as important. Often it will be mundane things that are seen as important. These actions do not cost a lot in terms of time or money, but they make a difference in families' lives.

Services that are currently offered as alternatives to home care may also require a shift in focus. Alternative services should meet the needs of families. Yet, services were often developed without input from families. Instead, they were based on the professional view of what families should require. There seems to be a critical need for professionals to work with families to develop appropriate services that are of acceptable standards. Reassessment of services and creation of acceptable alternatives deserve a high priority from both practitioners and policy makers.

Currently, the health care system focuses on providing institutionalized, acute care to individuals who present with acute episodes of illness. Yet, many illnesses are chronic. Both acute and chronic care are important, but the emphasis has been on acute care. Currently, non-acute services, such as respite care, are provided in a patchwork fashion that varies depending on where a family lives. Moreover, patients live at home and manage their illnesses within the context of their families. Not only does the person's illness affect the family, but the family has an effect on the illness. The importance of families and their incorporation into care plans seems to warrant some acknowledgement by care delivery systems. In addition, care that is provided from a family-centred perspective must also be integrated in a seamless fashion within the many components of the health care system. Integrated systems provide a continuum of care delivered by health care providers with the appropriate education and expertise, and allow for more comprehensive care. Resources can then be allocated to best meet the needs of the individuals, families, and communities within a given area.

Education of health professionals focuses primarily on ill individuals as the unit of treatment. Emphasis is placed on curing a particular illness. Other aspects of an individual's life, including the family, are often viewed as superfluous to the required treatment. Few health care professionals learn how to provide family-level care. Many are uncomfortable in discussing sensitive issues, such as sex, finances, terminal illness, or death. They shy away from talking

about feelings and reactions. Yet, these and other psychosocial issues are often critical areas for families. In addition, the organizational set-up of current health care, such as tight scheduling of physician visits, reduces opportunities for asking the kind of questions that are necessary for family-level care. Learning about a family's biography and providing family-level care take time and specialized skills. Neither of these are in abundance within the current system. However, professionals can learn ways of providing family-level care in basic and continuing education courses. Collaboration with colleagues and professional support groups can also be used to improve one's expertise. Lack of time in a busy schedule is often cited as a barrier to meeting with a family. Yet, learning about a family and providing appropriate information in one initial, longer meeting may reduce the number and length of future visits because the family needs are addressed and the suggested information or interventions are useful for the particular family.

Collaborative relationships between health care professionals and families seem not to be the norm. Yet, families clearly desire partnerships with professionals. A necessary element of such a partnership is mutual respect, but only some professionals seem willing or able to accord respect to all families. Respect may not come naturally to everyone, but it can be learned. Critical self-reflection about one's attitudes and prejudices towards different types of families is one way of heightening self-awareness. Clarifying values and exposing prejudices may encourage professionals to show more respect to families who are different from themselves. Social, cultural, or religious affiliations are only some of the ways in which families may be different from their health care provider. Reflective practice around gender issues and societal expectations may also help professionals recognize stereotypes and gendered identities. Once these issues are acknowledged, then professionals may be more able to discuss them with families and colleagues. In addition, professionals who use critical reflection in their practice may be less likely to reinforce such stereotypes.

Mutual trust is related to mutual respect and is also necessary for collaborative relationships. Most professionals expect families to trust them. Yet, they seldom trust families to have ownership and responsibility for their illness. Professionals tend to view themselves as the experts and ignore the families' expertise. Collaboration implies sharing information and equal participation in assumption of responsibilities, work, and decision-making. Professionals who work in collaborative relationships with families can benefit from the knowledge that families have accumulated during their experiences with illness. Professionals can then use this knowledge, in conjunction with their own professional expertise, to suggest interventions that are most appropriate for the family. Knowing the family's circumstances before making suggestions increases the likelihood that the interventions will be useful and may increase family compliance with suggested treatments. In addition, when professionals are willing to learn from families, they gain new knowledge that they can share with other families in similar situations.

Collaborative relationships reflect sociobehavioural principles of partnership. Professionals can learn how to function effectively in collaborative relationships with families through educational experiences and role modelling. It is important to note, however, that attitudes and values are developed over a life-time. Efforts to change professionals' attitudes and values may meet strong opposition. Examining one's own beliefs can be a difficult and sometimes embarrassing exercise. Educational efforts must recognize this potential difficulty. Educators might consider encouraging open discussion among students and professional colleagues, but not making any judgement or censure. Instead, educational efforts could involve modelling of appropriate behaviours. Education that aims to teach respect for and develop trust in patients and families might begin early in professional training and continue as a thread throughout every aspect of training, including continuing education in practice settings.

Collaboration among professionals provides a continuity of care that is also extremely important to families. Many families require comprehensive services that are beyond the

expertise of a single professional. The knowledge and ability of multiple professionals is often needed. Effective interprofessional communication and co-ordination reduce family stress, promote family understanding of care, and provide continuity of care. Continuity of care is facilitated by having at least one person within the health care system through whom information can be co-ordinated. This person, or co-ordinator, needs to know the family well and have contact with each service used by the family. The co-ordinator provides an ongoing link to a system that is complex and often confusing for families. When this link is not present, families frequently perceive care as fragmented and unsatisfactory. A nurse may be particularly suited to this role, but only if power equality exists among professions.

A lack of collaboration between professionals contributes to the fragmentation of care. Professional differences, rivalries, issues of power and control, as well as a lack of understanding about the roles of other professionals interfere with collaboration. The issues and barriers involved in interprofessional collaboration warrant discussion if collaboration is to be successful. One place to initiate discussion about these issues is in educational facilities. Currently, professional education is delivered in ways that do not foster interprofessional collaboration. Each professional group, such as nurses, physicians, or social workers, learns about health care in isolation from health care colleagues. Educational programs could integrate health professionals in shared classroom and clinical experiences that are designed to breakdown barriers and encourage mutual respect and collaboration. In addition, instead of viewing collaboration as a sign that they lack knowledge, professionals might consider collaboration as augmenting the skills and expertise available to families. Practising professionals are obligated to put the family's needs first. If another professional can better provide the care that a family needs, then there should be no hesitation in referring the family to that health care provider.

One of the most important needs for families is the need for information. Families want information that is offered at a level they understand and in a manner that reflects openness,

honesty, caring, and respect. Professionals need to critically examine how they impart information. The experiential meaning of information is very important to families. Yet, professionals seldom acknowledge this meaning. Families appreciate being given information about resources, so they do not need to search it out for themselves. In addition, they suggest that written information, perhaps in the form of a summary letter, handbook, pamphlet, or resource manual, would be very helpful if given at the time of diagnosis because they could return to it whenever they had questions. Moreover, families find it stressful to be given too much information. They suggest that professionals slow down their pace to match that of the family. If professionals give suggestions on an ongoing basis, they can plant the seed of an idea and watch it grow when a family is ready. Professionals might consider exploring other ways in which they can improve their information sharing. Educational theory and practice could form a basis for changes in how information is imparted to families. For example, practitioners who learn through continuing education that most of what one says is communicated through one's behaviours and tone of voice rather than in the words one uses, may reflect on their own manner of communicating. This reflection, complemented by a new appreciation of education and communication theory, may lead them to act and sound differently the next time they deliver an illness diagnosis.

In addition, therapeutic communication skills, which include observing, interviewing, supporting, and listening, are valuable when working with families. These skills allow professionals to interact in helpful and sensitive ways, such as when telling parents that their child has a progressive, terminal illness. They encourage health care providers to recognize and acknowledge the experiential meaning that situations hold for families. In many cases, a lack of time, skill, or confidence in using therapeutic communication discourages professionals from practising their skills. Often, families are then referred to a psychologist or counsellor who displays these specialized skills. Yet, therapeutic communication is vital when working with

families in health care. It is the basis for developing the mutual trust and respect that is essential for successful collaborative relationships between families and health care providers. Like any skill, therapeutic communication skills can be learned and usually improve with practice. Formal education in classroom and practice settings will equip the professional with the necessary basic skills. Only implementation of these skills in practice will hone and enhance them, and will improve confidence in one's ability to use them effectively.

The best intentions of caring professionals who desire collaborative relationships with families and who strive to offer family-level care can be hindered by the physical setup of the institutions and community settings in which they work. For example, office space is often limited and there may not be a room large enough to hold a family conference. In addition, privacy is usually an issue in offices, hospitals, and outpatient departments. It is very difficult to discuss private and emotional issues with a family if other people can hear the conversation. Some of these issues might be alleviated if at least one meeting between professionals and family members was held in the family home. Another issue arises when an individual is hospitalized. Hospital rooms are often too small to accommodate a cot for a parent or relative who wishes to sleep with an ill child. Even when there is some space, both parents or the whole family usually cannot be accommodated. The message sent to families, therefore, is that they are not needed or wanted. There is an obvious need to design facilities that accommodate families. As the ones who interact most often with families, professionals might consider taking the lead in advocating for changes that bring families into their rightful place at the centre of health care.

Fatigue was an important component of families' experiences. Yet, little attention has been paid to the impact that fatigue has on families. Professionals receive little guidance from the literature regarding appropriate interventions. In clinical populations, fatigue can adversely affect one's quality of life. It is also correlated with increased illness uncertainty and reduced feelings of healthiness or well-being. Fatigue may have similar effects on family caregivers. While most

of the existing fatigue research has been conducted with non-caregivers, it offers some guidance for relieving the fatigue experienced by family caregivers. For example, respite care, especially at night, may prevent fatigued parents from making mistakes and may allow them time to sleep.

Another benefit of respite care that is seldom recognized by professionals is its role in reducing the need for directed attention. Voluntary or direct attention requires effort. Families require ways to reduce the need for directed attention. Involuntary attention that does not require sustained effort promotes the rest and recovery of directed attention. Professionals can encourage families to participate in activities that they enjoy and that will take their minds off the situation. Distracting or diversional activities help relieve fatigue. Simple restorative activities, such as walking or sitting in a natural environment, gardening, bird watching, or doing crafts or hobbies, also relieve fatigue. Respite care allows family members time away from directed attention. Yet, parents need not leave the house to benefit from respite care. When they are relieved of their constant vigilance, their need for directed attention is reduced and they can then rest. Families sometimes go into slow motion to reduce the demands for directed attention. Their ability to process information then slows down. Such families may need extra time to work through new ideas or suggestions until they finally understand them and no longer find them stressful. It is important for professionals to slow down as well to match the slower pace of families.

Physical exercise may be another way of relieving fatigue in families. Exercise has been used successfully by patients to both reduce fatigue and to decrease their perception of fatigue. The benefits of exercise in fatigued caregivers has not been explored. However, if professionals promote exercise, plus provide respite, they may enable families to increase their exercise and reduce their fatigue. Finally, cognitive-behavioural interventions to change families' perceptions and beliefs about exercise, activity, and fatigue may result in improved outcomes.

Research

The grounded theory of *navigating uncharted territory* received some support from the literature, although there were important differences between families where a child had an NLTI and those where the illness was potentially curable. However, remarks from colleagues at presentations of this work suggest that the theory holds promise for transferability to other areas, such as neonatal intensive care units. Further research is needed to determine the fit of this theory in both life-threatening and non-terminal situations. Moreover, the applicability of these results to culturally diverse populations must also be examined.

Family nursing science has been developing rapidly in the last decade. A strong foundation for family nursing science, however, is still being created. When the family is seen as the unit of care, then family relationships must be examined. There is a need to explore ways in which the family interactional approach can be implemented and evaluated. Research on family nursing interventions and outcomes is very limited. Such research has usually focused on individuals and not on the family as a whole. Family research needs to include all members of a family, including the children. The approach taken in this study - interviewing the family as a unit as well as individual family members - appears to be one successful method for conducting research at the family level. Further research could confirm this suggestion.

A variety of both qualitative and quantitative research methods and data analysis techniques need to be developed and tested for their usefulness at the family level. Neither qualitative nor quantitative research alone is sufficient to capture the complexity of family life. Instead, there is a need to examine the links between qualitative and quantitative research. Practitioners need to know both the processes by which families manage the experience and the outcomes associated with different subjective perceptions and management approaches. There is also a need to pay attention to gender in family research. At present, it is unclear how important gender issues are, but this current study raised issues for further research. Additionally, it is

important to explore if there are gender differences across cultural and socioeconomic groups. Finally, further research needs to explore the experiences of extended family members, such as grandparents, when a child has an NLTI.

There are a number of potential research questions that arise from this current study. For example, there is little available research about the link or interface between families and the health care system. Yet, relationships within the health care system played an important role in this study. Further research is needed, using both qualitative and quantitative methods, to examine family interactions with the health care system. Thus, researchers can gain a better understanding of the types of health care interventions that are most appropriate for families.

Some interventions that were suggested by this study could be implemented and evaluated. Families clearly valued ongoing contact with one person in the health care system. A study could be designed to evaluate interactions between a co-ordinator and a family. Outcomes could include families' satisfaction with care and their perception of how fragmented the system is before and after the intervention. An evaluation of whether involvement of a palliative care team makes a difference in families' lives is also warranted. The palliative care team could be introduced early in the experience for some families, and later for others. A control group that had no contact with a team might also be included.

None of the families felt that their child was a burden. Most reported receiving some satisfaction from caregiving. Further research is needed to explore the sources of satisfaction that make caregiving bearable or enjoyable. We also need to know more about how professionals can best use these sources to improve a family's quality of life. A number of individuals reported personal development because of their caregiving experiences. Growth of the family as a unit was also hinted at. Future studies could explore the concept of personal growth as an outcome of families' experiences. Studies could also be conducted to examine how families develop resilience to challenges.

The impact of fatigue on families requires further exploration. While there is increasing research on fatigue with people who are ill, research with family caregivers is very limited. The concept of *going into slow motion* raises issues about teaching and learning in illness situations. Moreover, the applicability of attentional theory deserves attention. Studies are required to determine ways in which the need for directed attention can be reduced and involuntary attention promoted. Intervention studies could then examine the effect of restorative activities on fatigue.

The complex relationships between fatigue, depression, and attentional performance need to be unravelled in order to more fully understand the phenomenon of fatigue. Qualitative research regarding the feelings associated with fatigue, and the interventions that relieve fatigue would also contribute to better understanding of this phenomenon. Since fatigue is multicausal, multidimensional, and subjective, measurement tools must be self-reported and reflect the multidimensional nature of fatigue. Currently, there are no suitable tools for measuring fatigue in family caregivers. Two existing tools for measuring fatigue in clinical populations, however, show promising psychometric evaluations (Piper et al., 1998; Schwartz, 1998b). Tools such as these may prove adaptable to caregiving populations in the future.

Conclusion

Families moved through a process of *navigating uncharted territory* as they lived with a child who was dying from a neurodegenerative, life-threatening illness. This process was initiated by a precipitating event, such as one parent recognizing a deterioration in the child's motor skills. The emotions of fear, uncertainty, and grief gave impetus to the process. These emotions were very strong in the beginning. They gradually declined over time, but increased with subsequent precipitating events. The process of *navigating uncharted territory* was characterized by four dimensions - *entering unfamiliar territory*, *shifting priorities*, *creating meaning*, and *holding the fort*. Parents used strategies within these dimensions to manage the physical, cognitive, and emotional work arising from the situation. These dimensions and

strategies were constrained and facilitated by four intervening conditions - *relationships with health care providers, availability of information, gender differences, and communication between parents*. In addition, the process of *navigating uncharted territory* occurred within the broader context of the *acute, curative health care system* and the *sociocultural environment*.

This grounded theory extends our understanding of family caregiving experiences. Important implications for changes in the way health care professionals interact with families and in how health care services are provided have been described. Finally, directions for future research are proposed. It is clear that families need guidance and support from professionals in a collaborative relationship that extends beyond the boundaries of hospitals and institutions. This research provides a basis for changing the way we care for families of children with NLTIs, and, in doing so, making a significant difference in families lives. Our world of health care must change so families like this one no longer struggle in isolation:

When people like us end up into your world, don't leave us alone. Just don't shove us out of the door and leave us alone...once we left [the hospital], that was it. That was IT. It was like they were throwing us to the wolves.

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Appendixes

Appendix 1: Family Member Information Sheet

Appendix 2: Consent Form: Adult Family Member

Appendix 3: Parental Consent Form: Ill Child

Appendix 4: Parental Consent Form: Sibling

Appendix 5: Initial Questions for Interviews

Appendix 6: Letter to Families Requesting Feedback on Draft Analysis

Appendix 1: Family Member Information Sheet

Family Member Number: _____

Demographic Data:

Age: _____

Gender: _____

Marital status: _____

Duration of marriage: _____

Occupation: _____

Household income: _____

Ethnicity: _____

Highest educational level (grade and # of years): _____

Relationship to child with progressive, life-threatening illness: _____

Child's Diagnosis: _____

Length of time since child diagnosed: _____

Details of Observations and Interviews:

Dates of observations: _____

Dates of interviews: _____

Risks:

There are no experimental procedures involved in this study. Any potential discomforts resulting from being observed and interviewed will be minimal in most cases. For some people, participation in this study may sometimes cause emotional discomfort.

Potential Benefits:

You will not receive any direct benefits from participating in this study. However, you may find it beneficial to discuss your experiences with the researcher. It is anticipated that the results of this research will provide information for developing supportive nursing interventions to enhance the quality of nursing care for families where a child has a progressive, life-threatening illness.

Monetary Compensation:

There will be no monetary compensation.

Confidentiality:

Any information resulting from this research study will be kept strictly confidential. All documents and audio tapes will be identified only by code numbers and kept locked in a filing cabinet accessible only to Mrs. Steele. Your name will not be used in the research reports. Information obtained in this study may also be used for educational purposes and research that involves secondary analysis of the observations and interviews, with the understanding that any additional research projects that use the observations and interviews will be approved by the appropriate ethics committees.

If you have any questions or concerns at any time during the study, please contact Mrs. Steele at the number listed above. You may also contact Dr. Davies if you wish.

If you have any concerns about your treatment or rights as a research participant, you may contact the Director of Research Services at the University of British Columbia, Dr. Richard Spratley at 822-8598.

I have read the above information and I have had an opportunity to ask questions to help me understand what my participation would involve. I freely consent to participate in the study and acknowledge receipt of a copy of the consent form.

Signature of Participant

Date

Signature of Witness

Risks:

There are no experimental procedures involved in this study. Any potential discomforts resulting from being observed and interviewed will be minimal in most cases. For some people, participation in this study may sometimes cause emotional discomfort.

Potential Benefits:

Your child will not receive any direct benefits from participating in this study. However, he/she may find it beneficial to discuss his/her experiences with the researcher. It is anticipated that the results of this research will provide information for developing supportive nursing interventions to enhance the quality of nursing care for families where a child has a progressive, life-threatening illness.

Monetary Compensation:

There will be no monetary compensation.

Confidentiality:

Any information resulting from this research study will be kept strictly confidential. All documents and audio tapes will be identified only by code numbers and kept locked in a filing cabinet accessible only to Mrs. Steele. Your child's name will not be used in the research reports. Information obtained in this study may also be used for educational purposes and research that involves secondary analysis of the observations and interviews, with the understanding that any additional research projects that use the observations and interviews will be approved by the appropriate ethics committees.

If you have any questions or concerns at any time during the study, please contact Mrs. Steele at the number listed above. You may also contact Dr. Davies if you wish.

If you have any concerns about your child's treatment or rights as a research participant, you may contact the Director of Research Services at the University of British Columbia, Dr. Richard Spratley at 822-8598.

I have read the above information and I have had an opportunity to ask questions to help me understand what my child's participation would involve. I freely consent / do not consent to my child's participation in the study and acknowledge receipt of a copy of the consent form.

Signature of Parent / Guardian

Date

Signature of Witness

Assent of Child Obtained: Yes / No

Risks:

There are no experimental procedures involved in this study. Any potential discomforts resulting from being observed and interviewed will be minimal in most cases. For some people, participation in this study may sometimes cause emotional discomfort.

Potential Benefits:

Your child will not receive any direct benefits from participating in this study. However, he/she may find it beneficial to discuss his/her experiences with the researcher. It is anticipated that the results of this research will provide information for developing supportive nursing interventions to enhance the quality of nursing care for families where a child has a progressive, life-threatening illness.

Monetary Compensation:

There will be no monetary compensation.

Confidentiality:

Any information resulting from this research study will be kept strictly confidential. All documents and audio tapes will be identified only by code numbers and kept locked in a filing cabinet accessible only to Mrs. Steele. Your child's name will not be used in the research reports. Information obtained in this study may also be used for educational purposes and research that involves secondary analysis of the observations and interviews, with the understanding that any additional research projects that use the observations and interviews will be approved by the appropriate ethics committees.

If you have any questions or concerns at any time during the study, please contact Mrs. Steele at the number listed above. You may also contact Dr. Davies if you wish.

If you have any concerns about your child's treatment or rights as a research participant, you may contact the Director of Research Services at the University of British Columbia, Dr. Richard Spratley at 822-8598.

I have read the above information and I have had an opportunity to ask questions to help me understand what my child's participation would involve. I freely consent / do not consent to my child's participation in the study and acknowledge receipt of a copy of the consent form.

Signature of Parent / Guardian

Date

Signature of Witness

Assent of Child Obtained: Yes / No

Appendix 5: Initial Questions for Interviews

Individual

Individual family informants were asked to tell the researcher about their experiences when a child has a neurodegenerative, life-threatening illness (NLTI). Both current and retrospective information was sought. A conversational tone was used throughout the interviews. As the interview progressed, questions were asked that reflected back the interviewee's own words. The interviews began with open-ended prompts such as:

- Tell me about your life since (child) was diagnosed with (an NLTI).
- What has this experience been like for you?

The purpose of this approach was to elicit the informant's perspective with as few prompts as possible. Other broad open-ended questions that guided the first interviews included:

- Tell me about a typical day. How does your day start? What happens next?
- Is a typical day today the same as it was a month (or year) ago? What is the same? What has changed?
- What has helped you care for (child)? Can you tell me about times when something or someone has made it easier.
- What has made it difficult for you to care for (child)? Have there been times that have been particularly difficult? Can you give me an example.
- What is life like for you now?

Family

In family group interviews, the focus was on the family unit. Therefore, questions were reworded to reflect the family experience. Families were advised that some of the responses in the family interview may be the same as in the individual interviews, but other responses may be different. Additional questions included:

- Tell me what you do together as a family. Is this different from before (child) was sick? What is the same? What is different? Why might that be?
- What is life like for your family now?"

Appendix 6: Letter to Families Requesting Feedback on Draft Analysis

May 20, 1998

Dear Mother and Father,

I know it was a long time ago that we met and I interviewed your family, but it's now "next spring/summer" and, as promised, I have started drafting the findings from my dissertation research study. You said that you would be interested in seeing some results, so that's what I'm sending you.

I want to start though by apologizing for any difficulty this letter or reading the findings may cause you. I have no way of knowing if your circumstances with Child have changed, so please forgive me if this is a difficult time for you.

What I've done is take over 600 pages of interviews and tried to understand what's happening as families live with a child who has a neurodegenerative illness. That means that I've looked for similarities in what families told me, as well as differences because it's important to try to understand the range of things that people do. Then, after many hours of hard work, I tried to figure out "the story" that captured how families live their lives. This means that the findings represent a composite picture of "a family" rather than any individual family. You may recognize yourself in it, but other parts might not apply. That's okay. What I'm looking for is to try and give a broad picture of what it's like GENERALLY for families, always remembering that we are all individuals and there will always be things that are special or different for each of us.

As you read the findings (if you want to read them - you certainly don't have to!!), you'll notice that the writing is a bit abstract and "academic." That's because it has to be for my dissertation. But, it still needs to be understandable, so I hope I've managed to make it readable for you. Let me know if I haven't. There may be some parts that I can't change because of the academic requirement, but I'll change anything I can. You'll see that I used lots of quotes throughout. Some of you may recognize yourself, or may think you recognize your words, but the names don't fit or the sex of the child is "wrong." I did that deliberately to try to protect confidentiality as much as possible, so it's not a misprint or me being stupid!

I would be really interested in hearing your comments about what I've done. Does this idea of "Travelling through uncharted territory" make sense to you? Have I got it "wrong" some place? Is there anything I'm missing that you think is important? You can put your thoughts on paper and send them back to me in the enclosed, stamped addressed envelope. Or if you want to talk with me or even be re-interviewed, then give me a call. I will probably call you in a week or two if I haven't heard from you just to touch base. Hope that won't be inconvenient for you.

Thanks again for all the help you gave me for this project. I still feel privileged that people like you were so willing to open their hearts and talk with me. Take care.