LIVING UNDER CONDITIONS OF SUSTAINED UNCERTAINTY

by

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ABSTRACT

Biomedical advances of recent years have made long-term survival possible for many children with life-threatening, chronic illnesses and raised the possibility of a permanent cure for others. Consequently, the illness experience for the families of these children has been significantly altered by changes in both the nature and scope of stressors that they must confront. In all potentially fatal chronic illnesses, sustained uncertainty has emerged as a major source of family stress.

The purpose of this research is to make analytically explicit the ways in which living under conditions of sustained uncertainty transforms the everyday life of families. Specifically, the conditions that create or increase uncertainty, the interactions that occur around issues of uncertainty, the strategies that parents use to manage uncertainty, and the consequences of living with sustained uncertainty are described.

A grounded theory method was used to analyze data from three sources: a) an existing longitudinal data set consisting of tape recorded and transcribed interviews with the parents of 10 children with cancer; b) conceptually and substantively relevant literature on uncertainty; and c) interviews with a cross-sectional sample of parents of 21 children with a variety of chronic, life-threatening illnesses.

In addition to diagnosis, the families varied with regard to age and sex of the affected child, the amount of time elapsed since the diagnosis, and the intensity of current medical therapy.

The analysis uncovered a process by which parents pass from a secure, taken-for-granted world to a reconstituted, uncertain world. Although uncertainty is a major source of perceived stress, under certain conditions certainty may be more stressful. The management of uncertainty, therefore, requires strategies to reduce, create, or maintain uncertainty in six interactive dimensions of daily life. These dimensions are time, information, awareness, social interaction, the environment, and the illness.

The implications for nursing practice include the recognition and minimization of those events and situations that trigger a heightened perception of uncertainty. In addition the parents' changing needs to maintain or dispel uncertainty should be supported.

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CHAPTER 1

THE RESEARCH PROBLEM

The contradictions experienced by families whose child has survived cancer have been described by Koocher and O'Malley (1981) as the Damocles Syndrome. Damocles was a courtier of ancient Syracuse whose king invited him to partake of a sumptuous banquet, but seated him beneath a sword that was suspended by a single hair. The moral lesson was that the riches of the king's office were tempered by the constant threat to his survival.

It is just this kind of ambiguous situation that the families of children who are chronically ill with a life-threatening condition experience daily. On the one hand parents are told to be grateful for the medical advances that hold the promise of cure, remission, or arrest of the disease. They are advised to treat their child normally and plan for the future. On the other hand, they are constantly reminded that their child is not normal by the need for frequent visits to the doctor, continual parental monitoring, daily medications and treatments, restrictions on the child's activities, as well as the presence of physical stigmata or labile physiological processes. They are cautioned against unrealistic hopes for the future and advised to live one day at a time.

The stress created by sustained uncertainty in chronic illness is a process that is only beginning to be systematically studied in nursing. Yet it is a phenomenon confronting an increasing number of families living in societies with advanced biomedical technology available to its citizens.

Statement of the Problem

The central problem of this research is to make analytically explicit the ways in which uncertainty becomes part of the fabric of everyday life in the families of children with a life-threatening, chronic illness. The conditions creating uncertainty will be addressed, as well as the interactions that occur around the issues of uncertainty, the strategies that are used to manage both day-to-day and long-term uncertainty, and the consequences of living with sustained uncertainty.

The focus on uncertainty as a major source of family stress emerged during the fall of 1984 when I had the opportunity to work as a research assistant with Dr. Ida Martinson. The work involved the analysis of interview data from a longitudinal study of the long-term effects of childhood cancer on families.

Of the various themes that were identified in the data, it became increasingly clear that, for the majority of parents, uncertainty was the single, most pervasive stressor that confronted them (Cohen & Martinson, 1988). For as long as the child survived (and for half of the families the child survived disease-free for the entire five year period of the study) uncertainty affected the family's day to day activities as well as their thoughts about and plans for the future. It altered communication within the family and influenced the parents' relationships with others in their larger social network, including relationships with members of the health care disciplines. Because sustained uncertainty was found to be central in the lives of families whose child had cancer, and because there is a growing body of literature that supports the proposition that all chronic illnesses share generic properties, it is hypothesized that sustained uncertainty is a logical consequence of any life-threatening, chronic illness.

At the present time there are no studies that have directly addressed the conditions under which the phenomenon of uncertainty in chronic illness varies, the effects of sustained or chronic uncertainty on family life, or the management strategies that parents use to cope with this stressor. Because nursing is a discipline whose practice is defined as "the diagnosis and treatment of human responses to actual or potential health problems" (American Nurses Association, 1980), these issues are an appropriate focus for nursing research. Research that defines and makes explicit parental responses to sustained uncertainty and links them to the conditions under which the responses occur, has the potential to contribute to nursing knowledge and revise or expand nursing practice. The resulting substantive theory would have the power to explain the phenomenon, enable the prediction of behavioral responses under varying conditions, and suggest therapeutic nursing interventions.

Purpose of the Study

The purpose of this study is to describe and explain: (a) the subjective experience of sustained uncertainty, (b) the conditions which heighten or lessen the perceptual awareness of uncertainty, (c) the properties of uncertainty that cause it to be experienced as stressful, (d) the strategies that families use to manage the stress related to uncertainty, and (e) the long term consequences for the family of living with sustained or chronic uncertainty. The specific aim is to generate a substantive theory that will account for family behavior under conditions of sustained uncertainty and that will enable nurses and other health professionals to generate testable hypotheses relating to the nature and timing of appropriate interventions.

Assumptions Concerning the Significance of the Study

The four assumptions concerning chronic childhood illness that underlie and give significance to this investigation are: (a) there is an increased prevalence of chronically ill children in this country; (b) the medical achievement of long-term survival of chronically ill children has changed the nature of the psychosocial stressors confronting their families; (c) there are universal or generic properties inherent in all chronic illnesses; and (d) in any life-threatening chronic illnesses, uncertainty is a major source of sustained stress and is a significant factor in understanding a family's behavior.

The first assumption rests on the fact that the United States has been a leader in the development of a medical technology that has raised the possibility of a disease-free survival for a small percentage of children with previously fatal illnesses and, at the same time, created a population of medically or technology dependent, long-term survivors of diseases that cannot yet be cured. As a consequence, a larger number of American families than ever before are living with a chronically ill child member.

The second assumption states that not only is chronic childhood illness confronting more families, but the illness experience for these families has been significantly altered by the changed nature and scope of the stressors that are inherent in the new technology. The generic properties of these stressors have been found to cut across many disease categories. When the illness is not only chronic, but also life-threatening, a heightened, pervasive, and persistent sense of uncertainty seems to be emerging as one of the most powerful sources of stress.

The relationship of stress to decrements in psychological, social and physical well-being has long been described by researchers (Dohrenwend, et al., 1982). Hansen and Johnson (1979), however, state that the study of stress as a process has too often been neglected in stress research, and that events are generally seen as being imposed on an individual or a family as a single, short-term stimulus rather than as a complex set of circumstances that has a history and a future. More recently, Pearlin and Aneshensel (1986) have described a stress process paradigm in which illness is viewed as an event that is capable of creating a cluster of consequent stressors that, once created, are each capable of constituting their own source of stress.

The third assumption claims that the diagnosis of any serious chronic illness in a child is just such an event. However, most of the research that addresses either the stress of having a chronically ill child or the family's response to this stress has two major limitations to a comprehensive understanding of the problem: a) the research tends to be categorical or disease-specific, and b) it is usually focused on the beginning or the end of the illness trajectory. Hence, there is a considerable body of literature on the impact of a specific medical diagnosis on the family, on the process of dying, and on the stages of bereavement. However, we know very little about what it means to a family to have their child survive without being cured and even less about the stressful effects of chronic uncertainty on the family. The fourth assumption proposes that the way in which families manage the stress of sustained uncertainty will have long-range implications for the health and well-being of all family members and for the quality of family life.

The following overview of chronic childhood illness will serve to organize the evidence in support of the assumptions listed above.

Incidence and Prevalence of Chronic Childhood Illness

Chronic illness in childhood has been defined as "a disorder with a protracted course which can be progressive and fatal, or associated with a relatively normal life span despite impaired physical or mental functioning" (Mattsson, 1972, p. 801). More recently, it has been defined as a condition that interferes with daily functioning for greater that three months in a year, causes hospitalization of more than one month in a year, or (at the time of diagnosis) is likely to do so (Hobbs, Perrin, Ireys, Moynihan, & Shayne, 1984). With communicable diseases under control and effective treatment readily available for most infectious and acute illnesses, chronic illness has become the major health problem of children in the United States (Haggerty, 1975).

The onset of chronic childhood illness may occur at any age and, with increasing frequency, the diagnosis is being made prior to birth.

The demographics of chronic illness in children vary in one important aspect from chronic illness in adults. In the adult, a relatively small number of diseases accounts for the majority of the chronically ill population. However, the percentage of children affected with any one disease (with the exception of asthma) is quite small and the number of diseases is extensive. For several of the more common childhood diseases, estimates range from 0.13 to 0.14 per 1,000 live births for hemophilia and Duchenne muscular dystrophy respectively, to 8.0 per 1,000 live births for congenital heart disease (Gortmaker, 1985).

There has been scant evidence of an increase in the incidence of chronic childhood diseases. However, the prevalence of such diseases has risen appreciably due to the extended survival of affected children made possible by the biomedical and technological advances of recent years. Over the past two decades it has become increasingly possibly to save birth-damaged infants, repair complex congenital defects, control the progression of life-threatening illnesses, achieve long-term remissions, and extend life by the use of mechanical devices that support vital body functions. These advances have accounted for the rise in the number of chronically ill children from an estimated 7 to 10 percent of the child population in the past decade (Mattsson, 1972) to between 10 to 20 percent today (Gortmaker & Sappenfield, 1984; Stein, 1983). Conservative estimates indicate that 6 million children in the United States have some degree of chronic illness or disorder and that of those, 1 million, or 1 to 2 percent of the total child population, have a <u>severe</u> chronic illness (Hobbs, Perrin, Ireys, Moynihan, & Shayne, 1984).

Long-Term Survival

Increased survival has not only led to an increase in the prevalence of chronic illness in children, but it has also created a population of adults with health problems not encountered among this group before. A few examples will demonstrate these changes.

Adults with cystic fibrosis were a medical curiosity only a decade ago. Today, however, adults account for 30 percent of the population known to the Cystic Fibrosis Foundation registry (Hobbs, Perrin, & Ireys, 1985). In the mid 1960's, 50 percent of children with cystic fibrosis died by the age of ten. Today 80 percent reach adulthood with a median survival age of 21 years for females and 25 years for males. Some large cystic fibrosis centers currently project the median survival to age 30 (Fenald & Boat, 1987; Huang, et al., 1987; Phillips, Bohannon, Gayton, & Friedman, 1985).

Of children born with tricuspid atresia, a form of congenital heart disease, 50 percent will die by six months of age, 60 percent by one year, and 90 percent by ten years if no palliative surgery is performed. With palliative surgery, 50 percent may survive to the age of 15 years. Until 1968 there was no definitive surgery to repair this defect. With the development of the Fontan procedure, the five year survival rate following surgical repair is projected to be 87 percent and may be accompanied by a significant improvement in the quality of life (deVivie & Rupprath, 1986; Humes, et al., 1987).

Without high technology intervention, the life expectancy for a child with Duchenne muscular dystrophy normally extends to the late teens or early twenties. Today, however, with the availability of portable equipment to provide mechanical ventilation, some men in their middle 30's are still alive (Gilgoff & Dietrich, 1985).

The increased life span of children with life threatening chronic illnesses has profoundly affected their families, the society in which they are growing up, and the health care delivery system that serves them. In 1971, for example, Hoffman and Futterman wrote that one of the most important tasks for the parents of children with leukemia "...involves maintaining investment in the welfare and future of the sick child while also preparing for his death...(for) despite their efforts, the child is virtually certain to die." These children were not expected to return to school, participate competitively with their peers, or continue with the normal developmental tasks of childhood. Professional advice to families was to live a day at a time, and nurses studied ways to help parents through the process of anticipatory grief.

Today the long-term survival rate for children with leukemia is 60 percent or better (Meadows & Silber, 1985), and the tasks required of parents, society, and health care providers are significantly different. Parents must be dissuaded from preparing for the child's death, while acknowledging the possibility of its occurrence. The community in which the child lives and society at large must find ways to integrate these chronically ill children and young adults into the mainstream of community life. And health care professionals must develop new strategies to support the child, the family, and the community for a prolonged and indeterminate period of time.

Generic Properties of Chronic Illness

The research on chronic diseases of childhood and the service programs that have developed from this research are primarily based on a categorical approach that emphasizes the unique characteristics of individual disease entities and organ systems (Pless & Perrin, 1985). This approach has encouraged considerable specialization and subspecialization among medical researchers and, by narrowing the research focus, has made the rapid advances in disease-specific diagnostic and therapeutic techniques possible. The categorical approach to the study of chronic diseases of childhood is, unquestionably, both necessary and valuable and it has served well as the organizational framework for medical knowledge.

Funding and delivery of health care, as well as the advanced preparation of most health care practitioners, remains on a categorical basis. Arguments for maintaining a categorical perspective include the physiological diversity of the diseases, the uniqueness of treatments, and the variation in the natural history and the ultimate, expected outcome of each disease (Burr, 1985).

The categorical approach has also been the model most frequently used by nurse researchers who have sought to explain how a particular disease impacts on a child and family and to discover the unique stresses and coping strategies that evolve as a result of that specific disease. The effect of such an approach is to ignore or understate the generic components of chronic illnesses in childhood and to inadequately address the needs that are common to all families.

In 1975 Strauss argued for a noncategorical approach to the study of chronic illness by identifying the salient features of all chronic illnesses. He described them as long-term, uncertain, intrusive, expensive, often comprised of multiple disease entities that necessitate great efforts at palliation or control, and requiring a wide variety of ancillary services. Similarly, Pless and Perrin (1985) have stated that when the views of parents, children and others outside the medical system are obtained systematically and analyzed, "it becomes clear that there are a limited number of difficulties frequently experienced by many, if not most, families who have a child with a chronic disorder. These analyses suggest that the difficulties vary only slightly from disorder to disorder or from family to family. If anything, the nature of the family, more than the nature of the disorder is likely to determine the frequency with which certain problems are experienced."

In a study that compared the psychological status of individuals in five medical diagnostic categories, Cassileth, et al. (1984) found that psychological adaptation was fundamentally independent of the specific diagnosis. In a similar vein, Felton, Revenson, and Hinrichsen (1984) reported that the use of specific coping strategies is minimally explained by the medical diagnosis. Stein (1983) has also concluded that there is probably more variation within than between diseases with regard to the experiences and problems that confront families.

In a landmark study, Jessop and Stein (1985) studied 209 mothers of children with 98 different diagnoses and found that "significant differences on social and psychological variables were found to relate principally to four dimensions of illness: the interference with daily functioning of the child, normal or abnormal appearance, the presence of surgical procedures and whether the family needs to accept changes in conditions" (p.997).

Any severe chronic illness in a child stresses and drains the child and every member of the child's family over an indefinite period of time. In terms of the psychological effect of the illness on the child, Shapiro (1983) reported that the specific illness may be less menacing than the familial response to the illness. Investigators have also failed to demonstrate that the severity of an illness is predictive of the response that the child or family will have to it. In fact there is some evidence that the less severely afflicted may be more psychosocially dysfunctional due to their marginal status and the increased ambiguity surrounding societal demands and expectations (Madden, Terrizzi, & Friedman, 1982; McAnarney, Pless, Satterwhite, & Friedman, 1974).

Regarding the psychosocial functioning of the family and its members, many severe chronic illnesses have identical consequences (Stein, 1983). Some examples are listed below.

Marital Stress

There is always an increase in marital stress reported but not, however, an increase in the rate of divorce. In fact, in some studies, between 13 to 25 percent of the parents reported that the illness brought them closer together and strengthened their marriage (Buchanan, LaBarbera, Roelofs, & Olson, 1979; Firth, Gardner-Medwin, Hosking, & Wilkinson, 1983; Peck, 1979; Phillips, Bohannon, Gayton, & Friedman, 1985; Sabbeth & Leventhal, 1984). Sabbeth (1984) suggested that chronic illness may have its greatest impact in the way that family members come to interact with each other.

Financial Difficulties

Financial strain is common in all but the most affluent of families. Perrin and Ireys (1984) report numerous surveys which confirm that regardless of the type and extent of insurance coverage that parents may have, out-of-pocket expenditures can be as high as 25 percent of a family's income. In addition to gaps in medical coverage, there are other expenses that are seldom covered by third party payors. These expenses include transportation to and from medical appointments (which are often at a medical center some distance from the family's home); food and lodging for parents who choose to remain with their hospitalized child; structural modifications that may be needed for the home or car; equipment, appliances or devices that are either essential for the care of the child or that simply improve the quality of the child's life; child care expenses for siblings when the ill child's needs require parents to be away from home; and, lastly, loss of income due to forced absence from work when the parent must stay at home to care for the ill child.

Excessive Demands on Time and Energy

Medical appointments, repeated hospitalizations, and home treatment regimens cause problems in time management and can lead to chronic fatigue or physical exhaustion for many parents. All families have limitations in time and energy. When one child is ill the realistic consequence is that these limited resources of time and energy are channeled primarily to the ill child and the welfare of healthy siblings may become compromised (Siemon, 1984). In almost every study of the impact of chronic illness on families, the lack of time for the healthy siblings is perceived by parents as a problem. Most parents are aware of and uncomfortable with this reality, resulting in varying degrees of parental guilt. Sibling jealousy may develop and is often accompanied by negative behavioral manifestations. As Siemon (1984) points out, however, siblings may be impacted in positive ways as well, with the illness calling forth an inner strength, and greater sensitivity, maturity, responsibility, tolerance, and altruism.

Recurrent Hospitalization

Hospitalization of the chronically ill child occurs frequently and has been reported in some studies to be a major source of stress (Phillips, Bohannon, Gayton, & Friedman, 1985; Simon, 1984). In a study of 35 families of children hospitalized for cancer or other long-term disabilities, Knox and Hayes (1983) found that hospitalization entailed the loss of the established parenting role. Ferraro and Longo (1985) suggested that treatment protocols in the hospital often fail to take into account previous, successful home management and devalue parental knowledge of the child's needs and responses to various forms of therapies. Hospitalizations also confront parents with negative comparison cases and may force upon them the awareness that children do, indeed, die of their illness.

Social Isolation

Social isolation from neighbors, friends, and extended family is common, and parents frequently find they have acquired a "courtesy stigma" whereby they sense the uneasiness or reluctance of others to be around them because of their child's illness. Chesler and Barbarin

(1984) interviewed 95 parents and 21 friends of parents whose child had cancer. They found that parents had difficulty "going public" with the news of their child's illness and suggested that this reluctance may have set the stage for ongoing difficulties that they and their close friends experienced in trying to seek or provide support. They also reported concern regarding the potential invasion of privacy or alteration in the prior boundaries of the relationship between family and friends and the uneasiness that this caused both parties. Sex role issues were found to be operating and it was felt that, not only did fathers have a more difficult time asking for, or being open to, support from friends, but their male friends could not easily provide this help.

Dissatisfaction with Medical Care

Families with chronically ill children are frequently dissatisfied with the medical care provided them, yet because of the limited facilities equipped to deal with the medical aspects of their child's illness, they are unable to seek care elsewhere. Lau, Williams, Williams, Ware, & Brook (1982) found that 76 percent of the mothers of chronically ill children interviewed in the clinic waiting room, expected that their physician would discuss psychosocial concerns with them during their visit. Only 29 percent of the physicians asked about psychosocial issues at all, and of those who did, they limited their questions to a very narrow field. The investigators attributed the mothers' dissatisfaction with their child's medical care to this unshared expectation.

Restricted Mobility

Geographical and occupational entrapment is a source of stress for many families (Stein, 1983). Parents often feel that they are unable to seek or accept new career opportunities if it entails relocating to an area that does not have adequate medical facilities, appropriate housing, a suitable climate, or if the move will mean a loss of their health insurance through their current employer (Buchanan, LaBarbera, Roelofs, & Olson, 1979; Salk, Hilgartner, & Granich, 1972). Mothers are frequently forced to leave their employment or give up their career plans in order to care for the ill child (Schuler, et al., 1985; Buchanan, LaBarbera, Roelofs, & Olson, 1979). Family outings, vacations, and other travel plans may have to be cancelled or curtailed, and the use of public transportation with the ill child may be impossible.

Parental Role Conflict

Technology has made it possible to provide sophisticated care in the child's home that was previously only available in the hospital, the clinic, or the laboratory. The possibility of home care and the reality of economic and social trends toward cost containment, deinstitutionalization, and mainstreaming, have combined to expand the caregiving role of the parent of the chronically ill child and have added new stresses to family life. Parents are now forced to assume many sophisticated monitoring functions, provide all of the required daily treatments, and make many critical medical judgments that were previously the domain of health professionals. In addition, they find themselves in the discordant position of being a parent in a society that both mandates the integration of the sick child into the community, yet stigmatizes the family for having such a child.

All of the foregoing psychosocial stressors may be considered generic properties of chronic illness in that they are relevant across diagnostic categories. Uncertainty, however, is perhaps the stressor most refractory to successful management since neither the most sophisticated technology nor an unlimited amount of financial and human resources can remove it or reduce its effects.

Uncertainty as a Sustained Source of Stress

Koocher (1985) reports that "uncertainty regarding the duration of the illness or its ultimate outcome is probably the greatest single psychological stressor for the patient with a life-threatening illness." Others have reported similar findings with a variety of chronic illnesses (Forsyth, Delaney & Gresham, 1984; McKeever, 1981).

In their study of 60 leukemic children and their families, Comaroff and Maguire (1981) found that the experience of uncertainty was one of the two major elements that characterized the effect of the disease. Wiener (1975) identified uncertainty as the core category that accounted for most of the variation in the social and psychological problems of living with rheumatoid arthritis. And Mishel (1981) found that of 100 adults admitted to a hospital, uncertainty about events occurring during the hospitalization, rather than the events themselves, accounted for their appraisal by patients as stressful.

Hansen and Johnson (1979) argued that the uncertainties introduced by change, regardless of the nature of the change, are among the most stressful qualities of a changed situation, while Parsons (1980) states that, "exposure to uncertainty is perhaps the most negative aspect of what many have considered to be the central feature of human life and action as distinguished from lower forms of living systems."

In their discussion of an ambiguous loss such as might be experienced by a family who has a physically absent or chronically ill member, Boss and Greenberg (1984) identified the construct of family boundary ambiguity as a variable that is appraised as being more stressful than the loss itself. Family boundary ambiguity was defined as uncertainty about the permanence of the loss of a missing member, the potential loss of an ill member, or the loss of a member that was anticipated but failed to occur.

Shalit (1977) suggested that ambiguity has the highest threat potential because it affects one's ability to accurately appraise a situation. Breznitz (1983a), on the other hand, stated that ambiguous information may also serve to reduce the perception of stress by allowing for a greater amount of disconfirmation. This is analogous to the phenomenon described by Waller, Todres, Cassem and Anderton (1979) who found that some parents prefer to appraise their ill child's future as unpredictable rather than as hopeless.

The uncertainty that arises from a child's life-threatening illness is multidimensional and is focused around a number of issues. Often there is etiologic uncertainty, which has been reported to substantially increase parental stress (Collison, 1980; Lippman-Hand & Fraser, 1979a; Meadow, 1968; Rothstein, 1980). By virtue of the fact that the illness is life-threatening there is existential uncertainty about ultimate survival or, if the disease is progressive, the rate of progression and length of survival. There is always uncertainty about the effect of the illness on the child's development and the quality of life that can be expected. The late effects of many types of medical and surgical interventions are only just beginning to appear among survivors and are often unpredictable. In addition to unknown physiological sequelae, there may be social uncertainties such as the child's future ability to compete scholastically, gain employment, obtain health or life insurance, marry, or bear children. There is the uncertainty that arises in the day to day life of the family as it tries to carry out its usual activities. Plans for vacations, family outings, or other social events are dependent upon the fluctuating health status of the ill child and may be made only tentatively, while ongoing medical expenses may make a family's financial base precarious and result in economic uncertainties. There is also the uncertainty that arises within the parents concerning their competency to adequately interpret their child's behavior to determine whether it is an indicator of a normal developmental process, such as a reduced rate of growth or a benign illness such as cold, or whether it represents a serious threat to the child's survival and requires immediate medical attention.

Uncertainty, then, may be considered a generic source of stress that transcends all chronic illnesses, but one which is appraised as most stressful when the illness is also life-threatening.

CHAPTER 2

APPROACH TO THE STUDY OF THE PROBLEM

The selection of the research question was guided by its relevance to the disciplinary matrix of nursing practice. However, the methodological approach to the study of the problem was determined by the paradigmatic assumptions that best reflect the dominant perspective of nursing science and the research question itself.

Paradigmatic Assumptions

A paradigm is a world view, a perspective, a set of metaphysical assumptions about reality. Although these assumptions guide a researcher's actions, they are often hidden, unquestioned, or unconscious.

The naturalistic paradigm that guided this research provides an explicit set of assumptions; one that is most congruent with the philosophy of nursing. Table 1 lists those assumptions and contrasts them with the scientific paradigm that has long dominated nursing research (Jacox, 1981: Watson, 1985).

Method

This study uses a grounded theory method to analyze data obtained from three sources: a) an existing 5-year, longitudinal data set consisting of tape recorded and transcribed interviews with the parents

Table 1

Competing paradigms of inquiry (Adapted from Lincoln & Guba, 1985).*

Assumptions About	Scientific Paradigm	Naturalistic Paradigm
The nature of reality	Reality is single, tangible and fragmentable	Realities are multiple, constructed, and holistic
The relationship of knower to the known	Knower and known are independent, a dualism	Knower and known are interactive, inseparable
The possibility of generalization	Time- and context-free generalizations are possible	Only time- and context-bound working hypotheses are possible
The possibility of causal linkages	There are real causes, temporally precedent to or simultaneous with their effects	All entities are in a state of mutual, simultaneous shaping, so that it is impossible to distinguish causes from effects
The role of value	Inquiry is value-free	Inquiry is value-bound

* Y. S. Lincoln and E. G. Guba, Naturalistic Inquiry, copyright 1985 by the publisher. Adapted and reprinted by permission of Sage Publications, Inc.

of a child with cancer; b) conceptually and substantively relevant literature; and c) focused interviews and participant observation with a cross-sectional sample of parents whose child has a life-threatening, chronic illness.

Grounded theory is a qualitative research approach that was developed and reported by Glaser and Strauss (1967) specifically for generating and testing theory. (See also Glaser, 1978; Strauss, 1987.) It is most appropriately employed in new areas of investigation where no theory regarding a situation exists, or in areas of prior theory development when one wishes to gain a new perspective or develop the theory further (Stern, 1980; Strauss, 1987). Grounded theory is a rigorous process of data collection and data analysis involving the use of induction, deduction, and verification. In this method of research, data collection and data analysis proceed concurrently.

Generative questions concerning a phenomenon of interest lead the researcher to selectively sample. In selective sampling, individuals or groups are chosen for the initial collection of data based upon the researcher's belief that they can provide information regarding the phenomenon under study (Schatzman & Strauss, 1974). Initially the data are subjected to an intensive, line by line scrutiny. This analytic technique is referred to as open coding and leads to the discovery, naming, and dimensionalizing of categories or concepts. The purpose of open coding is to orient the investigator to the full scope of the phenomenon under study and to suggest generative questions, potential sources for obtaining comparative data, and tentative hypotheses. As each new piece of data is coded, it is compared to the already generated concepts and categories, as shown in Figure 1 (Glaser, 1978).

Memoing is done throughout the research process, but the type of memo will vary in depth, specificity, and intent according to the requirements of the different stages of the research. Early in the research process, memos basically serve to record the investigator's thoughts, questions, and initial and tentative hypotheses concerning potentially fruitful areas for further investigation.

Axial coding begins fairly early in the analytic process and entails intense coding around one category at a time according to the

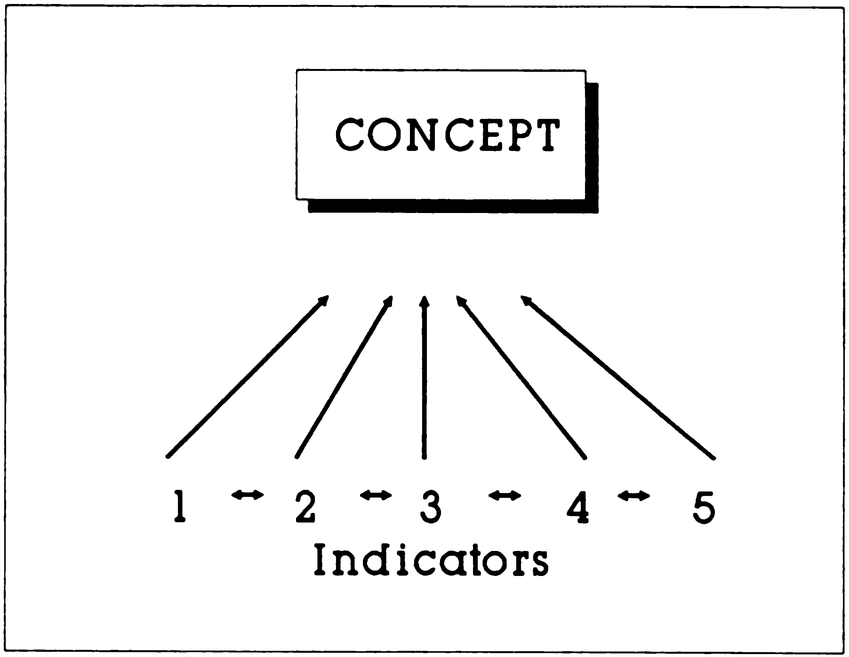

Figure 1. Operational diagram of the Concept-Indicator Model. From B. G. Glaser, Theoretical Sensitivity, 1978. Mill Valley, CA: The Sociology Press. Adapted and reprinted by permision of author.

coding paradigm of grounded theory as specified in Figure 2. This paradigm forces the researcher to focus on the conditions under which the category of behavior occurs, the interactions of the actors in the situation, the strategies used to manage the situation, and the consequences of those strategies.

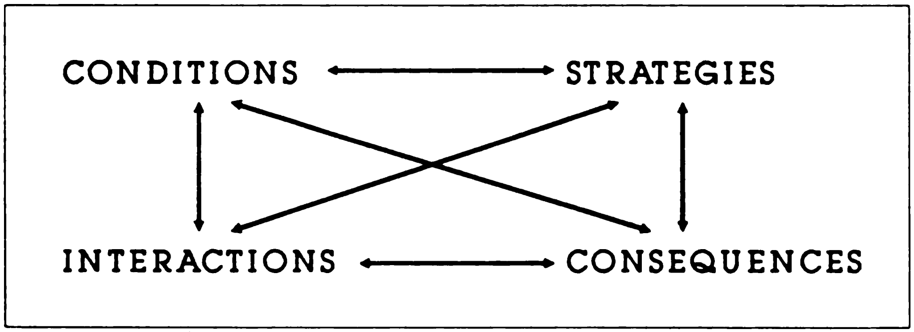

Figure 2. Coding paradigm of grounded theory.

As the analysis progresses, a core category will emerge that is central, appears with great frequency, relates easily to other categories, and has great explanatory power because it accounts for most of the variation in a pattern of behavior. Occasionally more than one core category may emerge. Following the identification of the core category, subsequent coding may be selective, rather than open. In selective coding, only those categories that relate to the core category are included in the analysis.

Theoretical memos are written in conjunction with data collection and coding in order to record the analyst's ideas about the properties of the identified categories and the connections between the categories and/or properties. Theoretical memoing is a necessary process that raises the level of analysis from a descriptive or thematic rendering of the data to a more conceptually abstract construction of theoretical hypotheses. These hypotheses form the framework for the emerging theory which is then tested through theoretical sampling.

Theoretical sampling is based upon analytic requirements, rather than upon *a priori* determinations of appropriate sources of data, as was the case in selective sampling. Unlike the sampling techniques used in quantitative studies whereby the researcher attempts to limit the variability of the object under investigation, grounded theory uses a process of theoretical sampling to achieve a full range of variation within a category. This technique assures a comprehensive representation of the phenomenon under study. Thus the inquiry will diverge, rather than converge, as subjects are chosen for their potential to bring out theoretically relevant differences as well as commonalities. In theoretical sampling, the appropriate sources from which to seek additional data are essentially dictated by the questions generated by the emerging theory. This constant comparative analysis of data in which new data are compared with the emerging theory, is the hallmark of the grounded theory method.

Integration of the emerging theory is accomplished by theoretical sorting of memos to reveal patterns of behavior, and by integrative diagrams which help to organize the relationships between the ideas that have been developed. Theoretical saturation occurs when continued data collection yields no new information about a category.

For a theory that has been generated by this method to be useful, it must be at a fairly high level of abstraction and be able to explain complex relationships or processes. This can only occur when the theory is conceptually dense; i.e., when it has has many concepts with multiple linkages (Strauss, 1987).

Theoretical Sensitivity

The quality of the product of the research is not only dependent upon the mastery of the techniques of grounded theory, as described above, but also upon the ability of the researcher to be "theoretically sensitive". To be theoretically sensitive is to be able to think about the data in theoretical terms (Strauss, 1987). In other words, to be able to go beyond a description of the phenomenon under study and to think theoretically about the concepts imbedded in the data.

According to Glaser (1978), entering the research setting with as few predetermined ideas as possible is the first step in gaining theoretical sensitivity. It allows the researcher to remain open to what is occurring without closing off potential sources of data or imposing premature structure to the analysis from extant theory or preexisting biases. Sensitivity is increased, however, by immersion in the literature dealing with the central variables once these variables have emerged from the data. Knowledge of the ways that the emergent variables have been conceptualized in other fields, as well as in one's own field, can generate ideas for the further collection of data and suggest potential concepts and categories around which to organize data.

It is the data that drives the literature search in grounded theory research and not the literature that directs the collection and interpretation of data. Chenitz (1986) states that there are many misconceptions concerning the purpose of a literature review in theory generating research. Unlike hypothetico-deductive or theory testing research in which the literature is used to suggest hypotheses or to operationalize an existing theory, grounded theory research always approaches the literature as a source of data in the form of written documents. These data are compared to the developing theory and are only utilized if there is an "emergent fit" between the extant categories and the generated categories (Glaser, 1978).

Design

A predetermined design serves the function of controlling the variance in the phenomenon under study, while an emergent design captures or expands the variance. The design of a grounded theory study must not be rigidly planned in advance. Rather it must emerge as the inquiry progresses and the theoretical framework begins to present itself (Lincoln & Guba, 1985).

This study progressed through a succession of three reiterative phases described below. The first phase was a secondary analysis of data from a longitudinal study of families who have or had a child with cancer. The second phase was a review of the literature on the concept of uncertainty. The third phase was the collection and analysis of data from a cross-sectional sample of families that have a child with a life-threatening chronic illness in one of five diagnostic categories. In the final rendering of the analysis, there was a continual shifting back and forth between all three phases.

Phase 1: Secondary Analysis of Longitudinal Data Set

In February, 1978, a longitudinal study was begun at a major Midwestern university medical center to determine the impact of childhood cancer on families.* The sample was nonrandom and consisted of a majority of the families of consecutively diagnosed children during an 18-month period beginning in February, 1978. Of the 49 families who were admitted into the study, 17 had a child who died during the study period, 16 had a child who was alive five years following the diagnosis, and the remaining 16 families were lost to long-term follow-up.

The families were invited to participate in the study during the child's initial hospitalization at the medical center. After written consent, the first interview was conducted with one or both parents within one to four weeks following the diagnosis and, for the families whose child survived, at yearly intervals thereafter for a period of

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* Funded by the American Cancer Society (Minnesota Affiliate), the St. Paul Foundation, and the Home Care Research Fund: Principal Investigator, Ida M. Martinson.

five years. If the child died, additional interviews were conducted at one, six, and twelve months post-death and were continued yearly thereafter until the end of the five year period.

After the initial interview, which was usually conducted in the hospital, parents were subsequently interviewed in their home. If distance or other events precluded a home visit at the appropriate interval, the interview was conducted by telephone, in the hospital, or in the outpatient department of the medical center. All interviews were tape recorded and later transcribed.

Using a semi-structured interview guide, the interviewer focused on the parents' perceptions of the events surrounding the diagnosis of cancer and the initiation of therapy, on current needs and concerns of family members, and on how the family was coping with this life-threatening or bereavement situation. The parents were also asked about the availability and quality of their social, psychological, and financial resources. The same interview guide was used for all contacts with the families, however, it was modified to include post-death experiences if the child died.

In 1984 a grant was awarded to Dr. Ida Martinson to support the analysis of the data that had been collected between 1978 and 1984.** At that time I joined the research team as a research assistant for the purpose of analyzing data from the interviews with parents.

From the larger sample of 33 families for whom longitudinal data were available, a subset of 10 families was selected for a thematic analysis. The decision to use only 10 of the data sets was based upon the time constraints imposed by the funding agency for the completion of

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** Funded by the California division of the American Cancer Society.

the report. The 10 families that were selected represented 10 of the most complete data sets over the five year period (see Table 2).

Table 2

Family Data Sets

Completed Interviews	Family Code Number									
	S1	S2	S3	S4	S5	D1	D2	D3	D4	D5
Initial	x	x	x	x	x	x	x	x	x	x
Postdiagnosis										
1 yr	x	x		x	x		x			x
2 yr	x	x	x	x						
3 yr	x	x	x	x	x					
4 yr	x	x	x	x	x					
5 yr	x	x	x	x	x					
Postdeath										
1 mo							x	x	x	x
6 mo						x	x	x	x	
12 mo						x	x	x	x	x
2 yr						x	x	x	x	x
3 yr						x	x	x	x	x
4 yr						x		x	*	x
5 yr						x				

Key: S, surviving child; D, deceased child.
*Withdrew from study.

Equal numbers of families with living and deceased children were included in the analysis in order to attempt to identify similarities and differences in the experiences and the responses of the two groups. Type of cancer, age and sex of the child, and other demographic factors did not enter into the criteria for selection for analysis. However, there were no significant demographic differences in the subset of ten families from the total population of families studied. Of these ten families, five had a child who died within four to twenty-three months following the diagnosis of cancer and five had a child who survived, disease-free, for five years after the diagnosis.

The transcripts of parental interviews were read several times in their entirety while simultaneously listening to the audio tapes. Substantive codes were generated from the data and organized into categories or themes. This initial analysis of data revealed many sources of stress that were common to all, or to a majority of the families following the diagnosis of childhood cancer (Martinson, et al., 1984).

Initially, ambiguity and uncertainty were identified as one of the eighteen themes that emerged from the data (Martinson & Cohen, 1988). During further analysis, uncertainty emerged as a core variable into which many of the other themes could be collapsed or to which they could be ultimately linked. This finding led to the next phase which was a comprehensive review of the literature on uncertainty.

Phase 2: Review of the Literature

In the second phase of this research, uncertainty, as the core variable under investigation, was reviewed from a number of disciplinary perspectives. The full review appears in Chapter 3. The following is a multidisciplinary overview of uncertainty that highlights the main focus of each of the disciplines.

Philosophy provided the context in which uncertainty was addressed as the antithesis of knowledge. Researchers in the field of econometrics or economic forecasting, reported extensively on decision making under conditions of uncertainty. Studies from the field of psychology have been primarily concerned with responses to event and temporal uncertainty. (Most often these studies were experimental and performed in a laboratory setting, frequently with subhuman species.) In the sociology and family literature reference to uncertainty was often included anecdotally to a study with some other major focus.

A number of studies and essays dealing with physician behavior in relation to the medical aspects of uncertainty in diagnosis, treatment choice and prognosis were reviewed. Nursing has begun to define and measure the properties of the concept in an attempt to develop a theory of uncertainty in illness.

Biographies written by the families of children with a life-threatening, chronic illness were included in the review of literature; however, these were used as an additional source of data to be included in the analysis rather than as a source of sensitizing theoretical concepts.

The concept of uncertainty from fields such as law, business, and politics was not reviewed at this time since these perspectives seemed less relevant to the present study.

Phase 3: Collection and Analysis of Cross-sectional Data

Because the Childhood Cancer study was not designed as a grounded theory study, the analysis was not concurrent with data collection and theoretical sampling was not employed. As a result, the issues relating to uncertainty were not systematically explored and were limited to the information that parents spontaneously volunteered. Nevertheless, these interviews were a rich source of data and allowed for some initial hypotheses from which to begin the third phase of the investigation. These hypotheses led to the construction of an appropriate interview guide for subsequent data collection (Appendix A).

In this phase, the categories from the thematic analysis were reorganized and recategorized around the question, "How do families experience and manage the stress of sustained uncertainty when their child has a chronic, life-threatening illness?" The tentative hypotheses thus generated were further developed by the processes of theoretical sampling and constant comparative analysis using the data obtained from a new cross-sectional sample.

The cross-sectional sample broadened the range of the phenomenon by including families of children with other chronic, life-threatening conditions in addition to cancer. The additional diagnostic groups included children with cystic fibrosis, congenital heart disease, and hemophilia.

Sample Selection

The majority of the families for this study were obtained from four pediatric specialty clinics at a large west coast medical center. Two families were obtained from a pediatrician in private practice. Single parent families, families residing more than 60 miles from the medical center, and families not fluent in English were excluded. Recruitment letters were sent to 29 families (Appendix B). Twenty-three families (79%) agreed to participate in the study; however, two families had to be dropped because of recurrent scheduling difficulties.

The final sample consisted of a total of 21 families whose child had either cancer (6), cystic fibrosis (6), severe congenital heart disease (4), hemophilia (4), or Lowe's syndrome (1). Two families each had two children with hemophilia. In addition to diagnosis, the families varied with regard to age and sex of child, time elapsed since diagnosis, and the intensity of current medical therapy. The ethnic/ racial composition of the sample was Caucasian - 76%, black - 7%, Asian - 10%, Spanish surname - 5%, and Filipino - 2%. Table 3 lists the characteristics of the children in the sample.

Rationale for sample selection. The fullest range of variability in a phenomenon is sought in developing grounded theory in order to guide the emerging theory (Chenitz & Swanson, 1986). The diagnostic groups were chosen because of their potential to maximize the variability of the dimensions of uncertainty as described below.

Only the families of children with cancer whose disease was in remission were selected for this phase of the study. When cancer is in remission it cannot be known with certainty whether the disease will ever recur; therefore, a condition of "event uncertainty" exists. In contrast, it is known with certainty that children with cystic fibrosis will die of their disease; however, the time of their death is uncertain. The average life span was previously noted (Chapter 1, page 8), but the range is from shortly after birth to the fourth decade of life. In this disease, a condition of "temporal uncertainty" exists.

Etiologic uncertainty exists for cancer and congenital heart disease, but not for cystic fibrosis and hemophilia. Cystic fibrosis and hemophilia are genetic. The former is transmitted by an autosomal recessive pattern in which both parents must carry the defective gene. Both male and female offspring may be affected. The latter is transmitted by an X-linked recessive pattern in which only the mother

Table 3

Characteristics of the Children in the Sample.

Sex	Age	Age at Diagnosis	Diagnosis	Birth Order
M	3.5 yr	6 weeks	CF	2/2
M	4.5 yr	Birth	Lowe's Syndrome	2/2
M	17.2 yr	1 day	CF	2/2
M	9 yr	5.5 yr	CA	1/2
F	9 yr	4.5 yr	CF	1/2
F	3.5 yr	2.5 yr	CF	1/3
F	14 yr	8 yr	CA	3/3
M	9 yr	3 yr	CF	3/3
M	5.7 yr	Birth	CHD	1*/2*
F	2.5 yr	3 mo	CF	3/3
F	10.5 yr	9.5 yr	CA	1/2
M	8.2 yr	8 mo	CA	1/4
M	4 yr	3.7 yr	CA	3/3
M	9.2 yr	5 mo	HEM	1/2 **
M	6 yr	2 wk	HEM	2/2
M	8.5 yr	22 mo	HEM	3/4 **
M	5 yr	Birth	HEM	4/4
M	8.5 yr	1 day	HEM	1/2
F	21 mo	8 mo	CHD	2/2
M	12 mo	3 day	CA	3/3
F	13 yr	10 day	CHD	7/7
M	6.5 yr	3 mo	CHD	1/2
M	17.7 yr	Birth	HEM	3/3

* Adopted **Siblings

CF - Cystic Fibrosis
CA - Cancer
CHD - Congenital Heart Disease
HEM - Hemophilia

carries the defective gene and only male offspring can inherit the disease.

Uncertainty concerning the late effects of treatment exists for cancer, severe congenital heart disease and hemophilia. These late effects cannot be predicted with any degree of accuracy for an individual child.

In the case of cancer, long-term survivors have at least a 10-fold increased risk of developing a second primary malignancy as a result of the radiation and, to some extent, the chemotherapy used to treat the original malignancy (Terracini, et al., 1986). Treatment with these agents may also cause learning disabilities, impaired physical growth, increased susceptibility to infection, and sterility.

The most serious treatment risk for children with hemophilia has been the transmission of the Human Immunodeficiency Virus (HIV) through contaminated blood supplies. New methods of preparing the concentrate used to treat the disease have virtually eliminated the risk of HIV infection for children who have been recently diagnosed; therefore, only families of children who have been receiving the treatment for six or more years were included in the sample. However, there is still a potential risk of infection from processing errors during preparation of the concentrate and parents are made acutely aware of this each time the child's blood is routinely checked for the presence of HIV antibody.

There is diagnostic uncertainty for those children who may have been previously exposed to the virus but who have not yet seroconverted (tested positive for HIV), and prognostic uncertainty for those children who are known to have been infected with the virus but who have not yet developed the disease.

The children with severe congenital heart disease whose families participated in this study either had, or were potential candidates for, a Fontan procedure. The Fontan is a surgical procedure that is performed for the purpose of correcting the hemodynamic abnormality that causes death in 90 percent of affected children by ten years of age. However, the risk inherent in the surgery can be as high as 15 percent and the long-term survival rate as well as the future quality of life is unknown because of the newness of the technique itself.

The remaining family whose child had Lowe's syndrome was included because it is a very rare disease about which little is known and which may or may not become life threatening.

Data Collection

Interviews were scheduled in the family's home at a time that was convenient to both the interviewer and the parents. Before beginning the interview, an informed consent was obtained from both parents (Appendix C). Children were not present during the interviews, nor were other household members.

Data were collected from parents by means of an intensive guided interview lasting 1.5 to 2.5 hours. The interviews were tape recorded and later transcribed. Both parents were interviewed concurrently except in three cases. In two instances only the mother was interviewed and in the third, only the father. Permission was obtained for follow-up interviews, as needed, to elaborate upon and verify the interpretation of the data. Participant observation provided a second source of data and these observations were recorded in the form of field notes following the interview.

Issues of Reliability and Validity in the Naturalistic Paradigm

The issue of the trustworthiness of research findings is as important in the naturalistic mode of inquiry as it is in the scientific mode. However, the criteria used to evaluate trustworthiness in each mode is a reflection of some basic and very important differences in the paradigmatic assumptions discussed earlier (see Table 1, p. 21). Guba (1981) identified four aspects of trustworthiness that evaluation criteria for all research must address: truth value, applicability, consistency, and neutrality.

Truth Value

While the truth value of scientific research is evaluated by how well threats to internal validity have been managed, the truth value of naturalistic research is determined by how credible the researcher's reconstruction of the multiple realities is to the constructors of the original multiple realities (Lincoln & Guba, 1985, Sandelowski, 1986). Credibility is enhanced by the use of several techniques: prolonged engagement, peer debriefing, negative case analysis, referential adequacy, and member checking (Lincoln & Guba, 1985).

In this research, the equivalent of prolonged engagement was achieved by the availability of the longitudinal data set from the Childhood Cancer study. With the exception of the initial interview, the tape recorded and transcribed interviews with parents were conducted almost exclusively by the same person over the five year period. This provided an opportunity to build trust and rapport between the parents and the interviewer and enhanced the quality of the data disclosed.

Peer debriefing "is a process of exposing oneself to a disinterested peer in a manner paralleling an analytic session and for the purpose of exploring aspects of the inquiry that might otherwise remain only implicit within the inquirer's mind" (Lincoln & Guba, 1985, p. 308). This was accomplished through regularly scheduled meetings with a grounded theory research group and additional sessions with Dr. Anselm Strauss.

Negative case analysis is the continuous revision and refinement of working hypotheses as more data become available, until a "reasonable" number of cases are accounted for. This was done throughout the analytic process. Lincoln and Guba (1985) state that if a hypothesis could be formulated that accounted for even 60 percent of cases, there would be substantial evidence of its acceptability. They claim that it is no more justifiable to expect to account for 100 percent of cases in naturalistic research than to expect a statistical finding significant at the .000 level in scientific research.

Referential adequacy involves "checking preliminary findings and interpretations against archived `raw data'" (Lincoln & Guba, 1985). Although archived raw data was originally intended to mean data that were collected by the investigator and stored "raw" to be retrieved later for the purpose of testing emergent hypotheses, in this study it refers to the use of published biographies, written by parents, about the events leading up to the diagnosis of their child and the subsequent patterns of living that evolved. In order to have consistency in the diagnostic categories between the archival data and the data obtained through interviews, biographical accounts of families of children with cancer (Gunther, 1949: Ipswitch, 1979; Lund, 1974), cystic fibrosis

(Deford, 1983), heart disease (Poole, 1980), and hemophilia (Massie & Massie, 1975) were included.

Member checks is the process by which data and interpretations are continually validated with the individuals from whom the data were originally obtained. It may also be done using subsequent interviewees who are asked to comment on the existing data or the researcher's interpretations. The latter method was used in this investigation. The investigator's reconstruction must be recognizable to the participants as an adequate representation of their own realities.

Applicability

The applicability of scientific research is referred to as external validity and is evaluated by the generalizability of the findings and the representativeness of the sample. Generalizations are truth statements that are time- and context-free within a given population. Naturalistic research, on the other hand, holds that phenomena are tightly bound to the time and context in which they are discovered and cannot be generalized to other situations *a priori*. If, however, it can be shown that there are essential similarities between two contexts, (i.e., if the conceptual linkages in one context are detailed and clearly specified and are later found to be the same in another context) it may be possible to transfer the findings from the first context to the second. Hence, "the naturalist does not attempt to form generalizations that will hold in all times and in all places, but to form working hypotheses that may be transferred from one context to another depending upon the degree of `fit' between the contexts" (Guba, 1981, p. 81). The possibility of transferability is enhanced by theoretical sampling and the density and specificity of the conceptual linkages.

In naturalistic research, the degree of transferability cannot be specified in advance as external validity can be by stating statistical confidence limits. Nor is it the original researcher's task to provide an index of transferability. Rather he or she has the responsibility to provide a rich data base with sufficient conceptualizations and specification of conceptual linkages to make transferability judgements possible by future researchers (Lincoln & Guba, 1985). The burden of proof of contextual similarity rests with those wishing to apply the findings of the original research.

Consistency

In scientific research reliability is based upon the ability of a test or instrument to yield consistently stable results every time it, or an alternate or parallel form of it, is administered to the same or comparable subjects. A reliable testing procedure is one in which the test administrator, scorer, or rater has developed consistent ways of giving the test and scoring or rating its results (Sandelowski, 1986). Reliability is a precondition for validity.

In naturalistic research, one does not expect to have consistent responses to the same instrument. In the first place, the instrument is the investigator and instrumental shifts are expected to occur as a result of developing insights on the part of the investigator. In the second place, given the assumption of multiple constructed realities, instabilities in responses from and between individuals are to be expected. Nevertheless, consistency is also required of naturalistic investigations, but is more accurately referred to as dependability. As Guba (1981) has stated:

> . . . for the naturalist, the concept of consistency implies not invariance (except by chance) but trackable variance--variance that can be ascribed to sources: so much for error, so much for reality shifts, so much for increased instrumental proficiency (better insights), and so on. (p.81)

Establishing an audit trail in order to permit a process check is one means by which dependability may be evaluated. The audit trail for this investigation specifies the process by which the data were collected and analyzed, and includes the verbatim transcriptions of the interviews, the field notes, memos, operational and theoretical diagrams. Establishing such an audit trail, makes it possible for an independent examiner who is familiar with the rules of the method to evaluate the process of inquiry and attest to the credibility of the product by confirming that it is supported by data.

Dependability does not imply that every (or even any) auditor who reads the data will independently arrive at the same conclusions as the investigator, only that by following the audit trail, he or she can affirm that the investigator's interpretations follow logically from the data and provide a plausible explanation of the phenomenon or process under study.

Neutrality

Within the scientific paradigm, neutrality refers to objectivity and implies freedom from investigator bias in the research process and product. Bias is seen as a consequence of methodological flaws. Rules of investigator conduct and research protocols are thought to maintain distance between the investigator and the subject and, thereby, help to reduce the threat of bias.

The naturalistic paradigm shifts the focus from investigator objectivity to data and interpretational confirmability, "requiring evidence not of the certifiability of the investigator and his or her methods but of the confirmability of the data produced" (Guba, 1981, pp.81-82). Leaving an audit trail and arranging for a product audit is one means by which the findings can be confirmed. The other is what Guba (1981) refers to as "practicing reflexivity." This means that the researcher intentionally reveals the underlying epistemological assumptions that lead to the formulation of a set of questions in a particular way, and to the presentation of the findings in a particular way.

Limitations of This Study

There are three major limitations to this study. The first is imposed by the constraints of time, resources, and the skill of the researcher. With such an extensive and rich data base, the research process could easily continue for several more years. With each return to the data, new generative questions arise and additional conceptual linkages become apparent, suggesting new areas to theoretically sample in order to create a more dense theoretical explanation.

The ability to think about data analytically rather than descriptively and the mastery of the techniques of the grounded theory method both require time to develop to their fullest. A first attempt cannot do full justice to the theory embedded in the data.

The second limitation to the study is the elite bias imposed by the sample. Overrepresenting data from an articulate, high status, well-informed group while underrepresenting data from less articulate, less informed, lower status individuals is a source of error that can limit the transferability of the emergent theory. Although socioeconomic status was not a criteria for inclusion in the study, most of the families were well-educated, middle to high income, and very articulate. The two families who did not fit this description did not provide sufficient data to indicate that uncertainty is currently a major source of stress in their life.

There are several plausible explanations for this variation, but with such a limited sample size, no interpretation is possible. One possibility is that families with limited financial and educational resources confront other powerful sources of stress that act as distractors, thereby diminishing the perception of uncertainty. One piece of evidence in support of this hypothesis is that one middle class family, in the midst of a separation pending divorce, when asked about the stress caused by the uncertainty of the child's status, indicated that, while it was a source of concern, it was being eclipsed by the fact that there were so many other changes going on at the present time.

A second possible explanation may be related to researcher effects. The social distance between the researcher and the participants may have created a pattern of communication that did not accurately convey the concerns of the participants.

A third possibility is the coincidental fact that the two families in question had children who had been diagnosed with cancer 3.5 years and 7.5 years prior to the interview, with no recurrence of the disease since the initial therapy. The length of time since diagnosis may account for uncertainty as a less prominent source of stress.

A third limitation concerns the boundaries imposed on the study by the groups that were included and excluded. Although the sample included a representative group with life-threatening, chronic illnesses, there was no attempt to include those with chronic illnesses that were not life-threatening. Nor were parents of children with acute life-threatening illnesses included. While the exclusion of these groups does not invalidate the findings of this study, it does limit the interpretation to the interaction of uncertainty and chronic, life-threatening illnesses.

CHAPTER 3

LITERATURE REVIEW

A review of the literature was undertaken to provide a sensitivity to those variables that helped to dimensionalize the concept of uncertainty. Only those that were of some help in organizing or interpreting the data are included here. In some cases they are referred to explicitly in the course of data analysis; in many instances their relevance is clearly apparent.

The Unknown and the Unknowable: Informational Deficit and Situational Ambiguity

When information is available but is not known to an individual, a condition of informational deficit exists. If the unknown information is not salient to the individual's present life situation, the lack of it does not create a subjective feeling of uncertainty and is, therefore, not defined as problematic. Should circumstances change and the unknown information later become salient in order for the individual to derive meaning from a situation, decide on appropriate actions, or make plans for the future, then the informational deficit becomes highly problematic. There are, however, recognized and effective information seeking strategies that will resolve this cognitive state of uncertainty. It is possible to make the unknown, known.

The realm of the unknowable consists of all ambiguous situations where information cannot be known with certainty by any individual. The unknowable, like the unknown, only becomes a stressor when what cannot be known becomes a central issue in one's life. But unlike the unknown, it creates a dilemma with no solution rather than a problem that can be solved through one's own efforts or with the assistance of others.

Ambiguity has been defined as characterizing a situation when information obtained from it is vague, unclear, incomplete, or has two or more possible meanings. It is also said to be present if the information contains inconsistencies, contradictions or has no apparent structure, organization, or is lacking in cues. Information may be considered ambiguous if it is conveyed in the form of a probability, and a temporary condition of ambiguity may be created if there is a long time interval between an event and the feedback of results (Lazarus & Folkman, 1984; Lorenzi, 1980; Mishel, 1981; Norton, 1975). Situations are likely to be perceived as ambiguous if they are novel, complex, or lacking in coherence (Budner, 1962). Ambiguity creates a state of uncertainty that Lerner (1980) describes as more diffuse than that which is created by informational deficit.

Tolerance of Ambiguity

There is some evidence from psychological research to suggest that the state of uncertainty created by ambiguous laboratory situations may be perceived as less stressful by some individuals than by others. It has also been reported that an individual's tolerance of ambiguity may demonstrate a degree of stability over time.

Harrington, Block and Block (1978) studied 120 preschool children attending nursery school. One item on the California Child Q-Set (CCQ), "Becomes anxious when the environment is unpredictable or poorly structured", was employed as the index of intolerance of ambiguity. Children who had been identified by a composite score of three nursery school teachers as intolerant of ambiguity at 3 1/2 years of age, were described by a different set of nursery school teachers one year later as significantly more intolerant of ambiguity than children who had not been so identified. The cross-time correlation of this index at time one and time two of .50 was uncorrected for attenuation leading the investigators to suggest a true reliability in the .70s or .80s.

Intolerance of ambiguity led to the avoidance of new or complex situations, restriction of attention to only a few elements of complex fields with little exploration, the imposition of unimaginative structure, and premature closure. Boys who had been described as intolerant of ambiguity at 3 1/2 and 4 1/2 years of age were still described as significantly more intolerant of ambiguity three years later, when they were in first or second grade. The were also described as more vulnerable to stress, more anxious, less competent and less resourceful. This was not true for the girls, although they were described as significantly more inhibited, less self-assured, and less assertive with their peers.

Intolerance of ambiguity was negatively correlated with the CCQ index of ego resilience which was described by items purporting to measure the child's ability "to react to the press of new and yet unmastered circumstances in resourceful, tenacious, but elastic ways." Therefore, the investigators claim that intolerance of ambiguity may be viewed as a facet or manifestation of the higher order construct of ego resiliency.

Budner (1962) conceptualized intolerance of ambiguity as a characteristic of the individual in which there is a tendency to interpret ambiguous situations as a source of threat. He demonstrated an empirical correlation between intolerance of ambiguity and socially relevant beliefs, behavior, personal value systems, and occupational choices. Specifically, the subjects intolerant of ambiguity tended to accept prevailing norms and values, believe in a divine power, and avoid exposure to conflicting value systems by favoring moral censorship. These behaviors were hypothesized to reduce the amount of ambiguity with which an individual has to contend and were all found to correlate with the measure of intolerance of ambiguity.

Norton (1975) found that persons who were highly tolerant of ambiguity, as measured by a 50 item measure of ambiguity tolerance, (MAT-50), tended to volunteer for undefined experiments and to use dramatization in small group problem-solving sessions more than low ambiguity tolerant people. They also tended to use different criteria to make aesthetic judgments than those who were less tolerant of ambiguity.

Although situational ambiguity is generally viewed as undesirable, under some conditions individuals routinely seek, rather that avoid, events with uncertain outcomes. The stock market investor, the test pilot, and the gambler are representative of those for whom uncertainty functions as a challenge rather than a threat. The greater the uncertainty of outcome, the greater their exhilaration.

Using sequential approximations to random shapes and English words and phrases as stimuli, Munsinger and Kessen (1964) reported an intermediate amount of cognitive uncertainty that was consistently preferred by subjects; one neither to low to cause boredom nor too high to be processed. Stimulus variability and cognitive structure were described as working in tandem to determine the amount of uncertainty an individual would prefer. This finding suggests that it may be possible to control some of the negative effects of cognitive uncertainty by decreasing the complexity of the stimuli or by increasing the individual's ability to derive meaning from the stimuli. Heinrichs (1984) reported that a moderate level of uncertainty mobilized cognitive effort and curiosity in accounting for the aesthetic reactions of interest and pleasure in human figure paintings.

One may conclude that in addition to seeking variety and novelty, people seek experiences that challenge their capacities. A challenge, then, may be thought of as any sought after or pleasurable experience where the outcome is uncertain.

Unpredictability and Uncontrollability

Uncertainty, as a function of unpredictability, has been studied extensively in stress research on animals, with the abundance of evidence suggesting that predictable, aversive events are preferred to unpredictable or unsignaled, aversive events (Lazarus & Folkman, 1984). In controlled laboratory experiments with humans, if subjects are given a choice between knowing when an aversive event will occur and not knowing, most would prefer to know (Monat, Averill & Lazarus, 1972). Evans, Phillips and Fearn (1984), however, found no simple relationship between the possibility of exerting control over an aversive event by information obtained from monitoring, and a subject's choice to monitor that information.

Individual differences not only make it difficult to interpret laboratory findings, but in many studies the desire to know has often been confounded with the desire for control. A study by Suls and Mullen (1981) examined the relationship between desirability of recent life events, perceptions of control over the events, and subsequent health among college students. The major finding was that uncertainty about one's control over undesirable events had a greater impact on increasing the incidence of illness than did having no control over undesirable events, and this difference was statistically significant at p<.01.

The lack of ecological validity of controlled laboratory experiments with short-term stressors of low salience has been a source of concern to researchers wishing to know how people react to aversive life events in naturalistic settings. Naturalistic studies have predominantly suggested that people want predictability in the events that have salience for them (Comaroff & Maguire, 1981; Okazaki, 1983; Roghman & Doherty, 1983). However, there are also those individuals who prefer not to know if an event will occur, especially if the prediction is expected to be adverse.

In a study by Mastromauro, Myers, and Berkman (1986), only 66 percent of the individuals at 50 percent risk for inheriting Huntinton's disease responded that they would avail themselves of presymptomatic genetic testing to determine whether they carry the defective gene. The most common reason given for wanting to be tested was to end the uncertainty of not knowing whether they would develop the disease. One-fifth of the sample even indicated that they would be relieved to know that they were gene carriers. Twelve percent of the respondents said they they did not want to be tested, with 53 percent of those expressing a preference to live with the hope that they would never develop the disease rather than risk certain knowledge that they are a carrier. A major gap in our knowledge is that we do not know the conditions under which people prefer to know or not know. Nor do we know the long-term consequences of uncertainty in matters that are highly salient for well-being.

Judgment and Decision Making Under Conditions of Uncertainty

"Coping with uncertainty or contingencies implies a capacity to decide among alternatives" (Parsons, 1980). A large number of psychological studies have focused upon the effects of uncertainty on an individual's ability to use sound judgment and make rational decisions when probability and risk are involved.

Probability

According to Montagna (1980), probability conditions are either mathematical, statistical, or subjective. As represented in Figure 3, certainty increases with increasing levels of probability. Mathematical probability represents those axiomatic types of absolute relationships which require no judgment and are based upon logic. It is, therefore, of little significance in the study of human behavior. Subjective probability, on the other hand, is said to occur when there is no known probability estimate upon which to base a decision. Statistical probability is based upon a relative frequency that is known or can be estimated.

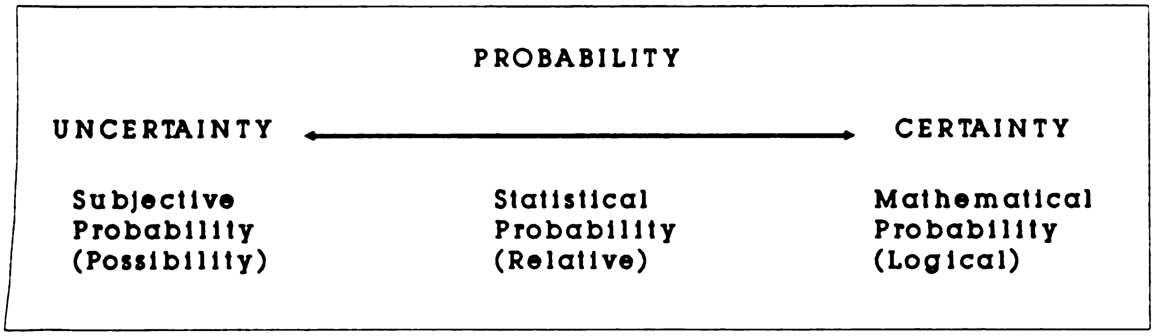


Figure 3. Montagna's description of the relationship of probability to levels of certainty.

Risk

Risk has been defined as a situation where actions lead to consequences with some known probabilities which are less than 1.0 (Milburn & Billings, 1976). Decision making under risk, therefore, has traditionally been defined as occurring when the probabilities are known or can be estimated, while decisions made under uncertainty reflect the individual's lack of knowledge of the probability of an event or the inability to assign a probability, perhaps because of the uniqueness of the event. One of the reasons for the confusion in the literature between the meaning of the terms risk and uncertainty has been the increasing popularity of the subjective theory of probability as opposed to the traditional view of probability as relative frequencies (Alpert, 1980). In a subjective theory of probability, individuals subjectively determine the size, distribution, and consequences of risk as a function of individual and situational determinants. Since all events are, thereby, assigned a probability the distinction between risk and uncertainty becomes unnecessary.

Models of Decision Making

In the classical model of rational decision making, the problem is clearly defined, all feasible courses of action are known, all possible consequences or outcomes are specified along with their attractiveness or aversiveness, and there is a probability attached to each action/consequence pair (Fischhoff, Goitein, Shapira, 1983). The more commonly used decision making model, however, has become the subjective expected utility model (SEU). A cornerstone of SEU thinking is that we live in an uncertain world and that all real decisions are made under uncertainty (Edwards, Kiss, Majone, & Toda, 1984). In this model the decision maker has subjective probability beliefs about the action/consequence set and he makes his choice of action based on his appraisal of the ability of his decision to maximize the utility (value) of the expected outcome (Milburn & Billings, 1976).

There is a growing realization, however, that despite its popularity, the SEU model also has deficits in its ability to describe the decision making process. Under conditions of uncertainty people are likely to accept the first decision alternative that "satisfices" (one that is seen as "good enough") instead of continuing to search for the optimal solution, or they may rely on past behavior, social norms, or the conclusions of experts. At times, they make only small, incremental decisions rather than a final one (Fischhoff, Goitein, Shapira, 1983).

Neither normative nor subjective decision theory provides an accurate description of actual human decision behavior outside of the laboratory. Under naturally occurring, stressful conditions it is not likely that an individual could know all feasible options or all consequences of his or her behavior. Real life problems are not structured in advance, nor do they exist in isolation. Rather they are part of an ongoing and developing process. As Lippman-Hand and Fraser (1979b) found, the decision to have a child, despite the presence of genetic risk, involved a process that included much more than simply coming to terms with the probability of the event and evaluating the burden of a negative outcome. It also included deeply personal interpretations of risk, a process of diffusing the decision making responsibility, and neutralizing the consequences by preparing for the worst.

Heuristic Biases in Decision Making

Recently there has been a shift in the focus of psychological research from the testing of models of decision making, to the study of the heuristics that individuals use in assessing probabilities and predicting outcomes or values, and the biases inherent in these heuristics. Tversky and Kahneman (1973) have reported that when people are faced with having to judge a probability in an uncertain situation, they employ a limited number of heuristics which reduce complex judgments to simpler ones. These heuristics, however, can lead to systematic and predictable errors. They have described, in detail, three of the heuristics that are commonly used: representativeness, availability and adjustment/anchoring (Tversky & Kahneman, 1974).

Representativeness

Representativeness refers to the degree to which people assess `A' to be representative of `B'. While this can be a very useful heuristic, it can also lead to several important biases, namely the neglect of base rates, insensitivity to sample size, misconceptions of chance, and misconceptions of regression. These are discussed in the following examples.

Tversky and Kahneman (1974) found that when subjects were asked to assess the probability that a personality description belonged to a lawyer or an engineer after having been told that the group from which the descriptions had been drawn consisted of 70 engineers and 30 lawyers, they based their assessments on the degree to which the description was representative of the two stereotypes, without regard for the prior probabilities or base rates of the two categories.

People also expect that a sequence of events generated by a random process will represent the true characteristics of that process without regard to sample size or to chance, and that deviations will self-correct by a deviation in the opposite direction. For example, a fair coin tossed three times and yielding three heads is judged more likely to come up tails on the next toss. Additionally, people tend to predict an outcome that is maximally representative of the input; therefore, those who scored highest on a prior test are predicted to score highest on subsequent tests.

Availability

Availability is the heuristic by which people assess the frequency of a class or the probability of an event by the ease with which instances or occurrences of these classes or events are called to mind. It produces biases because instances that are more easily retrieved or imagined are judged to occur more frequently. For example, when asked to judge whether more words had the letter `r' as the first letter or the third letter, most subjects incorrectly judged that the letter appeared more frequently in the first position because letters beginning with `r' were more readily available to recall than those in which an `r' was in the third position (Tversky & Kahneman, 1974).

Adjustment and Anchoring

Adjustment and anchoring is a heuristic in which estimates are made from an initial value and are adjusted to reflect that initial value. Subjects who were asked to estimate the product of 8x7x6x5x4x3x2x1 gave responses that were considerably higher than those subjects who were asked to estimate the value of 1x2x3x4x5x6x7x8. The difference in response reflects the different values obtained by the multiplication of the first few numbers.

Other Heuristic Biases

Researchers have identified additional heuristic biases. Borgida and Nisbett (1977) report findings similar to Tversky and Kahnman with regard to prior probability. In their study of how college students select courses, they found that face to face comments made by a few researchers posing as students who had taken specific courses were more influential in determining the students' choices than was the presentation of contradictory data taken from course evaluation forms that represented the opinions of an entire class that had taken the course. In this instance, base rates were ignored in favor of information from a few who were seen as representative of reasonable people with concrete, rather than abstract, information.

Similarly, Lippman-Hand and Fraser (1979a) found that in a genetic counseling situation, base rate information was not always perceived by couples as useful, although it could be recalled with ease when it was relevant (i.e., when couples had incomplete families). In lieu of utilizing base rates that limited the extent of uncertainty but did not eliminate it, couples chose to perceive the chance of having an affected child in a binary form, thus shifting the focus from risk or rate to outcome---the birth of a defective child would either happen or it would not.

Wallsten (1983), however, says that base rates <u>are</u> attended to when the following three factors are present: expertise of the judge, specificity of the information, and salience of the information. He has also noted that experts tend to abstract the basic principles of a problem, while novices represent the problem in terms of its literal features.

Bain's hypothesis, as cited by Cohen (1964), suggests another heuristic that biases decision making. The hypothesis states that people will be more convinced of the validity of a short series of items that is uncontradicted than they will be of a relatively long list that is contradicted by even a single item. For example, a treatment given to 5 patients with significant improvement noted in all 5 will be seen as more effective than a treatment given to 50 patients with 1 nonresponder.

In addition to making sizable and systematic errors in judgment, probability-learning research in laboratory settings has led investigators to claim that people simply do not have the cognitive schemata for effectively processing information and performing probabilistic tasks (Klayman, 1984). They are able to retain in short-term memory only a relatively small amount of information, they routinely make errors of logic in the simplest of syllogisms, and they learn simple combinatorial tasks very slowly. There is, therefore, a great disparity between the analytic demands of decision problems and the analytic capacity of the human mind (Connolly, 1980). Yet, in real situations, judgment and decision making are often noted to be quite efficient in probabilistic environments where large numbers of variables, complex interactions, and extensive uncertainties exist. As Connolly (1980) states, "We often seem to do much better than either our cognitive equipment or our knowledge of the world would justify."

Jungermann (1983) has placed these two views of human judgment and decision making capacity into two camps: the pessimists and the optimists. The pessimists, he says, believe that judgment and decision making under uncertainty show serious biases and errors which lie within the person and lead to violations of the principles of rationality. These violations consist of judgmental biases due to the reliance upon certain heuristics (which have been described above); representational faults, which seem to provoke the framing of decisions in a manner consistent with the way the problem was originally presented; and coping defects, as described by Janis (1982), where decision making behavior is perceived from a motivational perspective.

The optimists, on the other hand, claim that decision making is rational and it is the analysis of the decision making behavior that is defective. Their position is based upon three arguments. The meta-rationality argument states that the satisficing principle (see p. 53) is the rational choice when the costs of time and energy are taken into account. Because action is usually required within a finite period of time, discovery and consideration of all possible actions and consequences is inevitably and justifiably truncated. The continuity argument states that judgment and decision are moments in a continuous process and only appear biased or deficient if they are treated as discrete events. The structure argument claims that the assumption that the subjects share a common understanding of the problem with the experimenter is ill-founded.

Event and Temporal Uncertainty

Lazarus and Folkman (1984) distinguish between two types of uncertainty that may have some relevance for research in chronic illness: event uncertainty and temporal uncertainty.

Event uncertainty describes a probability estimate of the occurrence of an event, such as a relapse or a complication from therapy. The objective estimate, however, may vary considerably from a subjective one due to factors inherent in the person and the context in which the person arrives at the estimate. Individuals who have experienced a low probability event such as a life-threatening illness, have a heightened sense of vulnerability and may totally ignore objective, base rate information in predicting the possibility of a recurrence of the illness or the outcome of their disease. Temporal uncertainty refers to not knowing when an event is going to occur. It would be operating in those whose disease outcome is known to be fatal, but the time of death cannot be accurately predicted.

In experimental studies, the passage of time has been found to either heighten a threat or to allow time for reappraisal and the development of coping strategies (Lazarus & Folkman, 1984). As shown in Figure 4, using various measures of physiological and subjective emotional and cognitive states to assess stress, the majority of studies employing the temporal uncertainty paradigm generally report a gradual recovery even though the event may materialize at any time. The temporal certainty paradigm, on the other hand, shows a typical U curve with a period of recovery following the arousal caused by the threatening information, and a second gradual increase in arousal as the anticipated danger becomes imminent (Breznitz, 1983a; Monat, 1976 Monat, Averill and Lazarus, 1972). Because these studies were conducted in a controlled setting over a very brief period of time (perhaps only a few minutes to an hour) and since ethico-legal considerations prohibited the use of highly salient, aversive stressors, the results are not generalizable to situations of temporal uncertainty experienced in chronic illness. They are, however, useful in generating relevant questions for research.

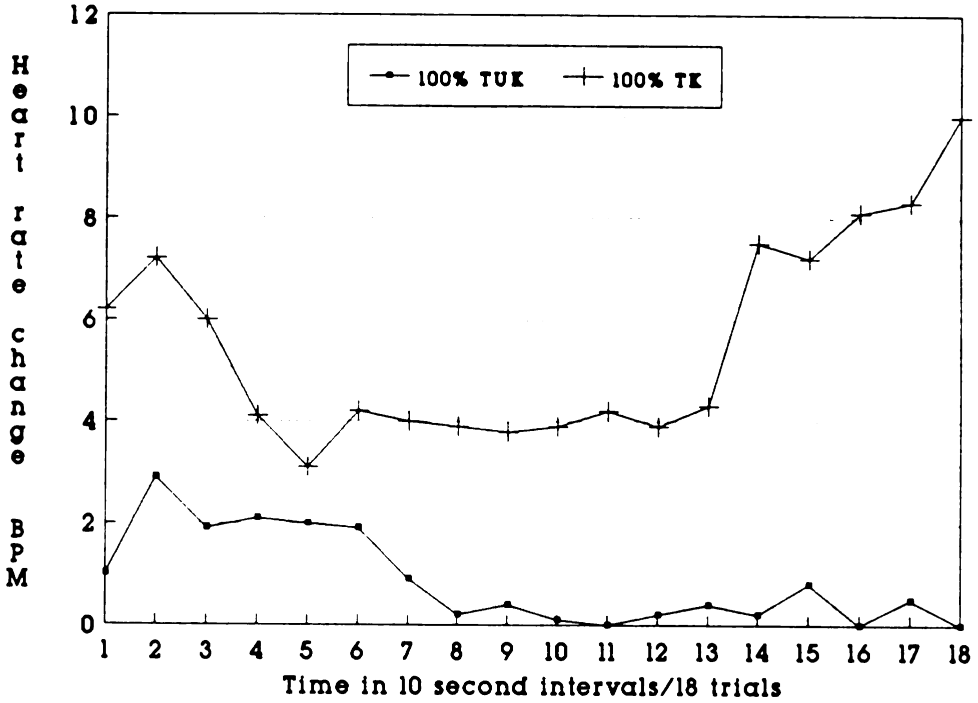


Figure 4. Represents the heart rate response during 3-minute anticipation of 100% certain shock when the time is either known (TK) or unknown (TUK). Similar curves were reported for skin conductance, relaxation-tension ratings and attention deployment. (A. Monat, J. R. Averill, and R. S. Lazarus, "Anticipatory Stress and Coping Reactions under Various Conditions of Uncertainty," Journal of Personality and Social Psychology, 24(2), 237-253. Copyright 1972 by the American Psychological Association. Adapted by permission of the author.)

Uncertainty in the Field of Medicine

Montagna (1980) states that professionalization is the process by which uncertainty is organized and controlled. It is done by the licensing of developed knowledge and the development and control of new knowledge. By licensure, a profession is granted exclusive control over the use of a body of knowledge.

The widespread intolerance for uncertainty is nowhere more apparent than in medical education and practice. Physicians are trained to make "right" decisions and do not manage uncertainty very well. Wolf, Gruppen, and Billi (1985) note five shortcomings in the physician's decision making process: 1) a bias toward positive and confirming evidence; 2) the primacy effects of initial information; 3) premature closure on a hypothesis; 4) inability to deal with probabilities; and 5) diagnostic conservatism. Their study of 89 first-year house officers revealed that even though the selection of a diagnosis is virtually determined by the information a physician seeks, only a minority selected data on which to base a diagnosis that were consistent with the competing hypothesis heuristic. This heuristic requires the consideration of each piece of evidence with respect to <u>all</u> hypotheses under consideration and is felt to provide the optimal strategy for diagnostic decisions.

Rhoden (1986) has identified the influence of cultural factors in the medical decision making process as it pertains to the treatment of infants with extreme prematurity. Doctors in the United States require a higher degree of prognostic certainty before terminating treatment than do British doctors, and they are also subject to many more nonmedical influences (such as malpractice litigation) in making their decision than either their British or Swedish counterparts.

Cohen (1983) suggests that physician training should include "uncertainty rounds", where clinical problems are presented that have no known or effective solution and students are supported in making rational decisions rather than intimidated into believing that there is a single right decision. He acknowledges, however, that having no right answer is likely to provoke great anxiety among physicians.

No matter how experienced a physician is, he cannot guarantee a patient that treatment will be effective because medicine is an inexact science and there is inherent uncertainty in both diagnosis and treatment (Frost, 1980). Yet the physician must, nevertheless, convey to the patient that he knows what he is doing. Katz (1984) states that physicians "will acknowledge medicine's uncertainty once its presence is forced into conscious awareness, yet at the same time they will continue to conduct their practice as if it didn't exist" (p.35). He suggests that physicians' disregard for uncertainty in doctor-patient decision making is due to a belief that some patients will not understand all of the options or choices available to them, and while others will not be benefitted by the disclosure of uncertainty. To quote Rothstein (1980), "Parents are confused by different options and generally do not have the knowledge to choose between different courses of action" (p.619). Katz (1984) also suggests that physicians may become immobilized if all the uncertainties of their practice are forced into conscious awareness.

In addition to uncertainty about the diagnosis, the choice and efficacy of various treatment modalities, the course of the disease and its ultimate outcome, and the late effects of treatment, uncertainty derives from many other sources. There is, for example, no unified opinion on what constitutes a disease or a pathologic finding in certain cases. What some practitioners may call hypertension, others call alterations due to the normal process of aging. Some diseases exist in medical texts only because advances in technology have permitted the identification of an aberrant anatomic structure or physiologic process. To many they are not considered diseases at all, but rather a normal variant with no known consequences for health.

An example of this diagnostic variance is provided by Eddy (1984) who reports that of 1000 eleven year-olds in New York City, 65 percent were found to have had a tonsillectomy. When the remaining children were sent to a group of physicians for examination, 45 percent were selected for surgery. The children who were not selected were then sent to another group of physicians and a similar percentage was, again, selected for surgery.

Physicians, uncertain about what diagnostic studies will give them the information they need most, tend to err on the side of excessive testing. The reliability of test results, however, is often dependent upon who performed the test, as well as upon patient factors, such as cooperation with the technician or compliance with the pretest preparation procedures.

Eddy (1984) claims that one way to fit a large problem into our minds is to lop off a significant portion of it. He says that when physicians are uncertain about the trade-offs that a given treatment entails, they often deal with only selected factors, such as the life-saving nature of the treatment and not the quality of life or economic issues that it raises.

The stress of uncertainty in medical practice is reduced for physicians by an educational system that fosters (and often demands) adherence to conformity and orthodoxy, by specialization which narrows the scope of the diagnostic information needed to practice, and by a professional tradition of action rather than inaction in the treatment of ambiguous disorders (Katz, 1984). There is also a growing interest in computerized medical data bases as a hedge against uncertainty (Cohen, 1984).

Calnan (1984) reviewed several interactionist studies of doctor-patient relationships and reported that uncertainty rarely became an issue during encounters between doctors and patients. Others, however, have not reported similar findings. Davis (1960) found that there were two types of uncertainty in doctor-patient interactions. The first was described as real uncertainty and applied in cases where clinical knowledge was lacking or ambiguous. The second he called pretended, or functional uncertainty. Functional uncertainty was operating whenever the staff became certain about a child's diminished prognosis, but the family was not informed of this. They were, "allowed to remain optimistically uncertain." The pretense of uncertainty on the part of physicians permitted them to invest less time and energy in encounters with the families and avoid confrontations with families who might be distraught by the prognosis. The pretense of uncertainty was felt to serve two other functions as well: it was believed that it would allow the families to maintain hope and that it would increase their compliance with the prescribed therapy.

Klenow and Dasilva (1980) reported a similar strategy among physicians treating dialysis patients whom they did not consider candidates for kidney transplantation. In order to maintain hope and compliance with the continuation of dialysis, the physicians allowed the patients to remain on the transplant waiting list despite the fact that they stated they would not perform the surgery even if a kidney became available.

In contrast, Waller, Todres, Cassem and Anderten (1979) reported four cases in which the physician gave parents an honest appraisal of a poor prognosis. In each instance, the parents rejected the information (and the physician) leading the researchers to conclude that parents would rather cling to an ambiguous prognosis than accept a hopeless one. They do not, however, recommend pretense but suggest sensitivity to the timing and context of the transmission of such information as a means of avoiding confrontation and the breakdown of the doctor-patient relationship.

Siegler (1975) coined the phrase "hanging of the crepe" to describe a strategy used by physicians in communicating a prognosis to families of critically ill patients. This approach offers the bleakest, most pessimistic outcome to families in an apparent belief that this will alleviate their suffering if the patient should die. By using this approach, the physician not only minimizes any uncertainty, but also maximizes his or her chances of being in a winning position if the actual outcome is better than that which was predicted.

If, as Fiddle (1980) claims, power can be demonstrated by the ability to transmit or produce uncertainty in others, then physicians may be seen as very powerful individuals. According to Light (1979), physician training includes training for five kinds of uncertainty: uncertainty surrounding the expectations and idiosyncrasies of teachers or mentors, uncertainty arising from the limitations of professional knowledge, uncertainty of diagnosis, uncertainty of treatment and outcome, and uncertainty of client response. In order to deal with the uncertainty of a client's response, he claims that medical trainees learn to gain control and establish a dominant relationship by increasing the client's uncertainty. "Trainees learn smoke screens and evasive replies that answer a client's question in form but not in content" (p. 316).

Yet actively foisting a state of uncertainty on others in order to avoid experiencing the helplessness of uncertainty oneself, is often a ploy of the powerless. While some physicians, undoubtedly feign uncertainty for the purpose of controlling the doctor-patient interaction or manipulating the likelihood of patient compliance, others who honestly attempt to convey real uncertainty often feel that patients place unreasonable demands on them for definite answers and then respond with unjustified anger when the physician is unable to comply. This type of interaction, repeated over time, causes some physicians to reach a point "where it is humanly impossible to display understanding" for the patient (Davison, 1984).

The Uncertainty of Chronic Illness

The uncertain status of most medical knowledge is, indeed, a source of stress for many patients and their families (Barbarin & Chesler, 1984; Cohn & Cohn, 1983). And in a society where the intolerance of uncertainty is high, patients have great difficulty learning to cope with the clinically unknowable. A study by Ben-Sira (1984) reported that although the physician's support was the most sought after by chronically ill persons, it was the least attainable in alleviating distress. When information _is_ available, it is frequently not offered. Instead patients may experience the withholding of information that is critical for the realistic appraisal of the impact of their illness and its effect on future aspirations and plans. Alternatively, the physician may provide an abundance of information but deny the complexity of the situation by reducing its management to simple rules of behavior such as a directing the patient to face only one day at a time, or by redirecting the patient's concerns from control of the disease to control of symptoms.

In his investigation of families of children with cystic fibrosis, Waddell (1983) described a process whereby confrontation with the epidemiological questions of the disease are deflected and the uncertainties that serve to destroy or inhibit hope are rendered inoperative. He refers to this process as "the ceremonial order of the clinic" which involves modifying the etiological question, holding out the promise of research, emphasizing the uniqueness of each case, making comparisons with less fortunate cases, and pointing out the lack of an alternative to the prescribed therapy.

Comaroff and Maguire (1981) found that most clinicians tried to deflect medical uncertainties and redirect the family's focus of concern away from the ultimate outcome and toward the immediate future. These attempts to deflect doubt about treatment efficacy and outcome were reported to exacerbate the effects of uncertainty. Longhofer (1980) states that by encouraging a one day at a time philosophy, "the patient is discouraged from invoking any future oriented thoughts and questioning and is thus forced to perceive his reality as unchanging" (p.126). Although a one-day-at-a-time strategy appears to be effective for individuals coping with severe, time-limited stressors, it may not adequately address the needs of the chronically ill who have a strong and ineradicable need for a knowable future.

The Effect of Uncertainty on Cognitive Appraisal

The potential for uncertainty to produce psychological stress is dependent upon cognitive appraisal. Cognitive appraisal is a continuous process by which an encounter is evaluated and categorized with regard to its significance for well-being. An event or situation is appraised as stressful to the extent that it taxes or exceeds an individual's ability to manage with his or her available resources.

Two interactive types of appraisal have been specified. Primary appraisal is an evaluation of what is at stake during an encounter and is categorized as benign-positive, irrelevant, or stressful. Stressful appraisals include those that indicate harm or loss has occurred, that a threat to well-being is present that may result in harm or loss, or that a challenge exists that has the potential for mastery or gain. Secondary appraisal is the evaluation of what can and might be done about the situation. Essentially, it is the appraisal of management strategies and their expected effectiveness in dealing with the stressor (Lazarus & Folkman, 1984).

Multiple factors within the individual as well as within the context of the situation determine how an event is appraised. Strongly held beliefs, motivations and values, as well as intellectual capacities, self-esteem and habitual or stereotypic ways of interpreting environmental cues are some of the person factors that account for variation in the appraisal of a given event (Lazarus & Folkman, 1984). Socioeconomic status, age, gender, marital status and location on the life continuum also play an important role in the way in which events are appraised (Pearlin, 1986). The timing of an event, previous experience with it, social role expectations, and the degree of situational clarity create the context in which appraisal takes place.

Davis (1963) described the model crisis experience of families whose child had contracted polio as a perceptual-interpretive appraisal process whereby a family passes from a secure state regarding its members, to one it perceives as grossly threatening. He outlined the following steps to the process:

1) Parents initially appraise the problem by applying an everyday, minimally threatening explanatory framework to the illness.
2) Increasing incongruities make it more and more difficult to continue applying this framework and parents become uncomfortably uncertain of their explanation.
3) Parents initiate action by consulting a physician.
4) A diagnosis is obtained.
5) Parents reappraise the illness as serious and dangerous.

In a similar vein, Featherstone (1980) described a process of increasing dissonance in the appraisal process in which the enactment of an inner dialogue takes place. One voice reassures the parent that there is no need for concern, while the other voice is disquieting and raises questions of serious abnormality. The dialogue continues only as long as each voice retains some credibility. Eventually, the reassuring voice is silenced when a definitive diagnosis is obtained. At this point diagnostic uncertainty is transformed into a more pervasive existential uncertainty. For these parents, their world has been fatefully and inexorably changed. They now live in a world where the worst is possible and they, more than others, are exquisitely vulnerable.

The Effect of Uncertainty on Interactions and Management Strategies

What people do to manage a stressful situation is a dynamic process, not a final achievement, and is oriented toward either altering the situation that is causing the stress, controlling or lessening the emotional and physical responses to it, or attempting to derive meaning from the situation by altering the perception of it (Lazarus & Folkman, 1984; Pearlin, 1983, Pearlin & Aneshensel, 1986). These three management strategies have been referred to as problem-focused, emotion-focused, and appraisal-focused (Moos & Billings, 1982).

The management of stress in families may be viewed as the product of a role set of interacting people (Pearlin, 1983b). The family provides the members with a repertoire of management strategies and an interactive context in which those strategies are accepted, encouraged, or constrained. Within the family, then, stress management is a process that is enacted by individuals, but the function that it serves and its success in reducing stress for any one member is very much influenced by the nature of the family's collective life--its themes, role sets and interactive patterns. Thus the responses of each person within the family may be either a resource to other members or a source of additional stress. Within the context of the family, a member's strategies are adaptive when they promote the maintenance of established patterns of behavior or encourage the negotiation of new patterns that will best satisfy the physical, social and psychological needs of each of its members, enhance the bonds between them, and enable them to participate in the mainstream of community life.

Breznitz (1983b) has reported that the duration of anticipation of a threatening event increases the amount of difficulty in dealing with it. It is, therefore, important to distinguish between acute, life-threatening conditions and those which are chronic when discussing the impact of uncertainty on coping. Many life-threatening conditions of childhood are acute problems that result from trauma or infectious diseases, and while much uncertainty may surround the initial diagnosis and prognosis, that uncertainty is usually resolved relatively quickly and unambiguously. The outcome for a child with a chronic life-threatening illness, on the other hand, may not be ascertainable and families of such children may experience an infinite period of unrelenting uncertainty.

In his study of 20 patients undergoing bone marrow transplantation, Longhofer (1980) identified the simultaneous, paradoxical messages of hope for recovery and fear of death as characterizing the communication from medical staff to patients. Under these conditions he states that patients often ". . . become obsessed with efforts to find hidden meanings in every conceivable message (or) . . . give in to the paradoxical injunctions by passively accepting all communication without reference to meaning" (p. 132). Mason (1985) also reported that for patients with diabetes, uncertainty was increased by the physician's exhortation to "lead a normal life" because they were also given information incompatible with normality: that the disease was serious, incurable, required daily medication, and careful control of diet and exercise.

Uncertainty can have an immobilizing effect on the family because the strategies that may help a family cope with an event's occurrence may be incompatible with the strategies that are needed to cope with the event's non-occurrence. Projected plans or thoughts may be immediately countered by opposing ones, thereby effectively neutralizing any anticipatory efforts at strategizing (Lazarus & Folkman, 1984).

Stress that is brought about by etiologic or existential uncertainty is least likely to be successfully managed by information-seeking strategies that pursue definitive answers where none are known to exist. Yet the need for complete knowledge has been reported to be so compelling that parents will often try to extract an answer by consulting a series of physicians, by invoking metaphysical explanations, or by the construction of a reality from group norms or social comparisons. Comaroff and Maguire (1981) report a particularly descriptive example of one parent's attempt to reduce uncertainty by this means.

> As a life insurance broker, he attempted to collate relevant data on the course of the disease from all available sources--doctors, paramedical personnel and other parents. He devised a multifactorial model of risk and survival for the population at hand, against which he plotted his son's prognosis (p.123).

Phipps (1985) describes the sense of uncertainty and vulnerability that parents experienced during a subsequent pregnancy after having lost a child through stillbirth or neonatal death. Parents tried to manage the uncertainty concerning the outcome of the current pregnancy by becoming hypervigilent, playing out negative scenarios, and taking a very task-oriented approach to the pregnancy while withholding emotional engagement.

Several investigators have reported that hope plays a pivotal role in managing the uncertainties of chronic illness (Forsyth, Delaney & Gresham, 1984; Klenow & Dasilva, 1980; Sandelowski, 1987; Wiener, 1975).

Hope, as distinct from denial, is a cognitive refocusing on the optimistic possibilities of a situation rather than the threatening probabilities. Denial may or may not be a concurrent strategy. The focus of hope will vary with the phase of the child's illness trajectory and the medical resources that are available. Initially parents may hope that the diagnosis is incorrect. Later, as the diagnosis becomes an accepted reality, they may hope for a cure, an arrest or remission, an absence of complications from therapy, a relief from suffering, or a peaceful death. In the face of medical uncertainty, hope may be easier to maintain.

Summary

The review of the literature identified a number of concepts or variables that were theoretically sensitizing in developing research questions for this study and in analyzing the data during and after collection. These concepts included the distinction between informational deficit and situational ambiguity (the unknown and the unknowable), and the variations within and between individuals to tolerate ambiguity, unpredictability, and uncontrollability. The review included the process of decision making under conditions of uncertainty when probability and risk are significant factors, and how the use of common heuristics is used to attribute causality, form judgements, and bias decisions.

A distinction was made between event and temporal uncertainty as two potentially productive dimensions of the phenomenon that have not been adequately explored in naturalistic research to determine their significance.

The literature on the uncertainty that is imbedded in the practice of medicine suggests that this may account, at least in part, for the nature of the interactions that take place between doctors and patients. Strategies that doctors use to deflect uncertainty have varying consequences, both for the patient and the doctor.

The literature on the effect of uncertainty upon cognitive appraisal of a situation or event, and on subsequent interactions and management strategies was considered to be at the very core of this research.

CHAPTER 4

UNCERTAINTY AND THE ASSUMPTIVE WORLD: AN OVERVIEW OF THE CONCEPTUAL RESEARCH PERSPECTIVES

> To predict the historical future is one of mankind's oldest yet unfulfillable desires.
> Jahoda, 1980

The Ubiquity of Uncertainty

Uncertainty has always been a condition of human existence and attempts to reduce uncertainty are deeply embedded in the history of the human race. If the various strategies to manage uncertainty are looked upon in terms of their generic properties, it is evident that these strategies have remained surprisingly constant throughout the ages, although many of the contingencies that give rise to uncertainty have changed with the changing times.

Primitive man lived in an extraordinarily precarious and uncertain world with few means at his disposal to understand natural phenomena or defend himself against the hazards that surrounded him. Events that could not be traced to their natural causes were seen as the uncertain consequences of uncontrollable and unpredictable forces. Any object that was conspicuously evident during a triumphant or tragic experience, a successful or failed venture, a period of prosperity or adversity, was accorded a particular causal significance in having effected the outcome and was thereafter perceived as a harbinger of good or an omen of evil. Man either sought or avoided such objects in an attempt to gain some sense of mastery or predictability of the events that governed his life.

It was in this atmosphere that primitive religion was born as man began to seek not only linkages between events, but reasons for them (Dewey, 1960). Those objects and events that had come to signify meaning or portend the future, became recognized as the instruments through which the gods exercised their power over man.

If man could not directly control the world in which he lived, he could, perhaps, influence the behavior of the gods. Ceremonial rites, ritual acts, supplication, and sacrifice were performed to appease those gods who had the power to destroy or bestow great benefit upon the petitioner. Security, however, came not only to those who were able to make successful appeals to the deities and thus control their circumstances, but also to those who were able to gain knowledge of the future or understand the meaning of current events. To reveal the intentions of the gods was to know the future. To this end their secret plans were sought through divination. Messages that foretold the future or explained the obscure were believed to come from the gods through natural phenomena such as dreams, birds, comets or eclipses, and were interpreted by the oracles, prophets and soothsayers. Often phenomena were not natural at all, but artificially and purposefully devised to penetrate the unknown. Opening a sacred book at random and treating as prophecy the line on which the eyes rested, or casting stones into water and divining from the sounds that were produced, are but two examples (Cohen, 1964).

The pervasive existence of uncertainty in human societies throughout history is evident from the fact that expressions for varying degrees of certainty can be found in most languages (Zimmer, 1983). That uncertainty has always been a source of stress, is indicated by the accounts dating back to antiquity of man's efforts to know the unknown. Modern day correlates of these ancient attempts to manage uncertainty are easily recognizable, though widely variable, in today's cultures. Many will become apparent in the analytic rendering of the data in Chapters 5 through 7.

The Dimensions of Uncertainty

Uncertainty is a multidimensional concept that varies in degrees of magnitude, intensity, and saliency--from the overarching existential issues concerning the creation of life and the mysteries surrounding death, to the inconsequential contingencies and probabilities that are the substance of everyday life. The source of uncertainty may come from within the individual as he questions his beliefs, values and self-worth, or it may arise from conditions in the environment that are perceived as novel, ambiguous, lacking in information or posing a potential threat to well-being. The perception of uncertainty may be disparate or congruent with reality. Uncertainty may be an overwhelming source of stress that accompanies uncontrollable or seemingly random events, or it may be a sought after challenge that acts as an antidote to boredom or as a means to test one's mettle. The scope of uncertainty may be total or partial concerning an event or interaction; it may be time-limited or persist indefinitely. It may affect either critical or noncritical areas of one's life and have either serious or minimal long-range consequences. Lastly, the experience of uncertainty may be socially shared or biographically unique.

The Awareness of Uncertainty

It is fortunate that we are never fully conscious of all of the uncertainties that are woven into the fabric of human existence for if we were, that awareness would be incapacitating. As Featherstone (1980) says:

> Most of us were raised to keep our eyes on the road ahead. We went to schools that justified long hours of dreary, mind-dulling drill and memorization by citing our future needs for skills. We sat quietly, walked in straight lines, and completed history assignments because these activities led to good grades and good jobs. As adults we continue to work and plan for a brighter tomorrow (p. 29).

If the events in our lives were seen as the unpredictable consequences of uncontrollable forces, what purpose would there be to plan for the future or engage in activities that did not offer immediate rewards? The degree to which we are able to envision our future as being continuous with and under the influence of our biographical past and present, determines the amount of coherence in our everyday life. The assumptive world in which we live provides that sense of continuity or coherence, thereby reducing the amount of perceived uncertainty.

The Construction of the Assumptive World

We are born into a sociocultural reality--a prestructured set of shared beliefs, values and customs that characterize a distinct world view. By taking for granted these socially and culturally prescribed interpretations of reality, we are able to categorize our experiences, narrow our choices in making decisions, come to know our world as having order and stability, and thereby reduce the amount of uncertainty that we must face daily (Cherlin, 1980; Kiev, 1976). The sociocultural environment not only reduces the amount of perceived uncertainty by providing standards and circumscribing expectations for decision making, but it is also important in determining how we respond behaviorally to the experience of uncertainty (Wright and Phillips, 1980).

Sociocultural reality is augmented by biographical reality. Biographical reality derives from our own unique experiences in the world and our interpretations of those experiences. Our biographical reality may either confirm or invalidate small or large portions of the intersubjective world of socially and culturally shared meanings.

Sociocultural and biographical realities together form the assumptive world from which daily life is constituted and enacted. The passage of time brings about change which produces fluctuations in the amount of convergence and divergence between our biographical reality and the sociocultural reality in which we live. The greater the degree of convergence between the two realities, the more likely we are to understand and be understood by others in the context of our daily lives.

The assumptive world is based upon the commonsense knowledge of everyday life and includes everything we know or think we know about reality at any given point in time (Parkes, 1971). It provides us with trustworthy recipes for appraising a situation and determining appropriate conduct, thus making what could be problematic, unproblematic.

The assumptive world consists of a relatively stable _cognitive_ world of accumulated knowledge, values, beliefs, and expectations, and an _action_ world of predictable events, routines, behaviors, and social relationships (See Figure 5). It encompasses the total set of assumptions that serves as a frame of reference for interpreting past and present experiences and for anticipating things to come. The assumptive world is the world that we take for granted.

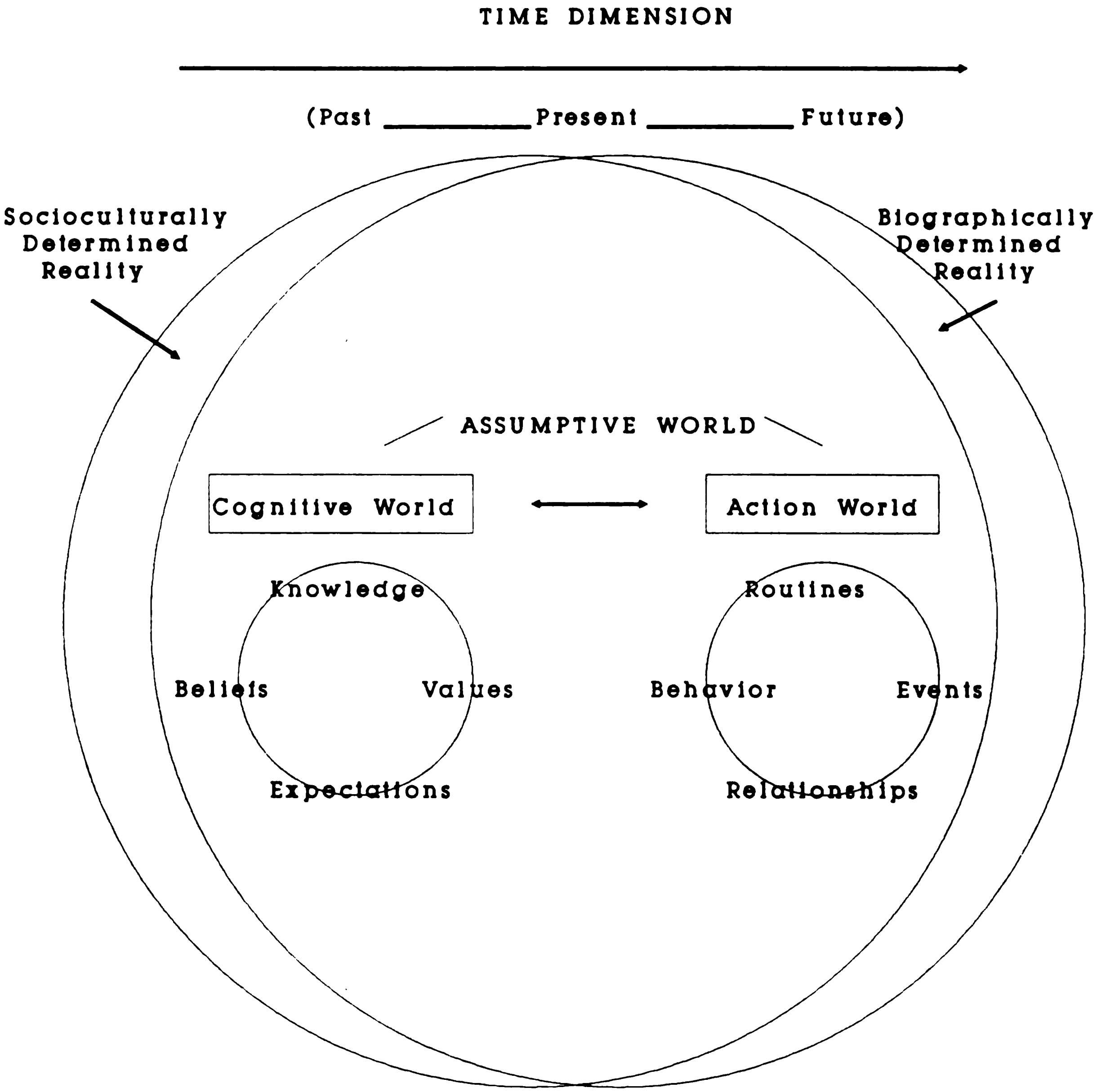


Figure 5. Construction of the assumptive world. Degree of convergence of sociocultural and biographical realities fluctuates over time but remains relatively stable. Time is perceived as continuous.

"To take the world for granted beyond question implies the deeprooted assumption that until further notice the world will go on substantially in the same manner as it has so far; that what has proved to be valid up to now will continue to be so . . ." (Schutz, 1970, p. 80). Even though we may know that imbedded in the reality of everyday life are unforeseeable and perhaps sinister possibilities and that there are no certain outcomes, we are able to suspend doubt and continue to conduct our lives as if reality were ordered and prearranged. Despite the weight of evidence that tells us that we live in an uncertain world, we simply do not expect the unexpected.

An experience that emerges in the course of daily life will not challenge our assumptive world as long as it can be categorized as "familiar". This means either that it is recognized as being another instance of a previous experience, or that it can be interpreted as "similar enough" to a previous experience to be considered familiar. It is the nature of our reality that we seek to interpret new or novel experiences so that they do not threaten the existing assumptive world. Neither will an experience pose a threat to our assumptive world if it is deemed to be lacking in pragmatic relevance to us at the time.

An experience becomes problematic when it has salience and attempts to place it in the realm of the familiar are unsuccessful. Or an experience may be initially perceived as familiar but come to be defined as "strange" and, therefore, problematic if our commonsense knowledge of everyday life fails to adequately account for its various dimensions. In the extreme case, an experience may be so oppositional to the existing reality structure that it causes a near-total transformation of the assumptive world. A near-total transformation is one in which the individual experiences an abrupt discontinuity with the past and the anticipated future and, as Berger and Luckmann (1966) explain, "switches worlds":

> The old reality, as well as the collectivities and significant others that previously mediated it to the individual, must be reinterpreted _within_ the legitimating apparatus of the new reality. This reinterpretation brings about a rupture in the subjective biography of the individual in terms of "B.C." and "A.D."...following the formula "Then I _thought_...now I _know_"....In addition to this reinterpretation _in toto_ there must be particular reinterpretations of past events and persons with past significance....a radical reinterpretation of the meaning of these past events or persons in one's biography (p. 147).

The Pre-illness Assumptive World of Parents

By the time individuals become parents, they have accumulated a sociobiographical set of assumptions about child bearing and child rearing that forms the unintrusive background for the daily enactment of family life. Once in the parental role, they develop and continually improve upon their sense of parental competency regarding their perceptions, interpretations, and actions as caretakers of their child. Although these "parental" assumptions constitute only a portion of everyday reality, they are interactive with the whole of it. Each new biographical experience and each new item of socioculturally transmitted information has the potential to expand, confirm, or alter parental cognitions and actions. A particular experience or piece of information may be sought by the parents or it may intrude on their life unexpectedly.

The current stock of knowledge which constitutes the preconstructed set of parental assumptions provides the mechanism for making parenthood and family life less problematic. By creating a sense of continuity with the past and future, the assumptive world allows parents to routinize many decisions and thereby reduce the amount of perceived uncertainty in everyday life. Many of these assumptions are brought into conscious awareness only when they are questioned or challenged.

CHAPTER 5

THE PREDIAGNOSTIC PERIOD

> When I was twenty-five, a friend told me that her brother had been killed in a motorcycle accident...She said, "Nothing bad had ever happened to me before...." What could she mean? Bad things happen to everyone--even to the luckiest people. Two years later I learned that my newborn son was blind; on that day I remembered her words. Now I understood them. I knew that nothing bad had ever happened to me before.
>
> Featherstone, 1980

The diagnosis of a severe chronic illness in a child constitutes a profound challenge to the assumptive world of parents. When that illness is also life-threatening, it brings about a near total transformation of that world. The perceptual-interpretive process by which parents pass from a relatively stable, taken-for-granted, pre-illness world to a reconstituted world of chronic uncertainty is the first analytic objective. The assumptions that underlie this process form the basis of the emergent theory.

The Prediagnostic Stages

The prediagnostic period consists of three non-recurring stages that follow the apprehension of the child's illness cues by the parents: the Lay Explanatory (Stage 1), the Legitimating (Stage 2), and the Medical Diagnostic (Stage 3). Although there is no evidence to suggest that these stages are perceived as separate and distinct by parents (except perhaps retrospectively), demarcating them for theoretical purposes is a useful heuristic in the development of a grounded theory since each stage accounts for significant variation in parental patterns of behavior.

There are three salient dimensions of each of the prediagnostic stages that must be made explicit. The first, which may not be immediately apparent, is the analytic focus. The focus is not on the parental unit, but rather on the process by which parents pass from the preconstituted assumptive world to the reconstituted uncertain world. The parental unit provides the setting in which the stages of the process vary and each family will vary the process to a greater or lesser degree.

Time, the second dimension, is quite evident by the fact that there is a beginning and an end to each stage with clearly discernible transition points between stages. This dimension is variable rather than fixed since the transition from one stage to the next is contingent upon the occurrence of a specific set of conditions or a critical event rather than a fixed time interval. A given stage may last for only a few minutes or for several years.

Sequencing is the third important dimension. It is fixed rather than variable. As the data frequently demonstrate, one or more of the stages may be omitted entirely, but the progression from one stage to the next can occur only in the forward direction. That is to say, for example, that the process may move directly to stage 3 from the pre-illness period (or from stage 1 to stage 3, omitting stage 2) but it cannot move backward to an earlier stage (see Figure 6). Stage 1, therefore, is not to be interpreted as the required starting point for families. Such a view would tend to deny the multiple realities of human existence and trivialize the process as well. Rather it should be viewed as the operational representation of one possible starting point in the diagnostic process.

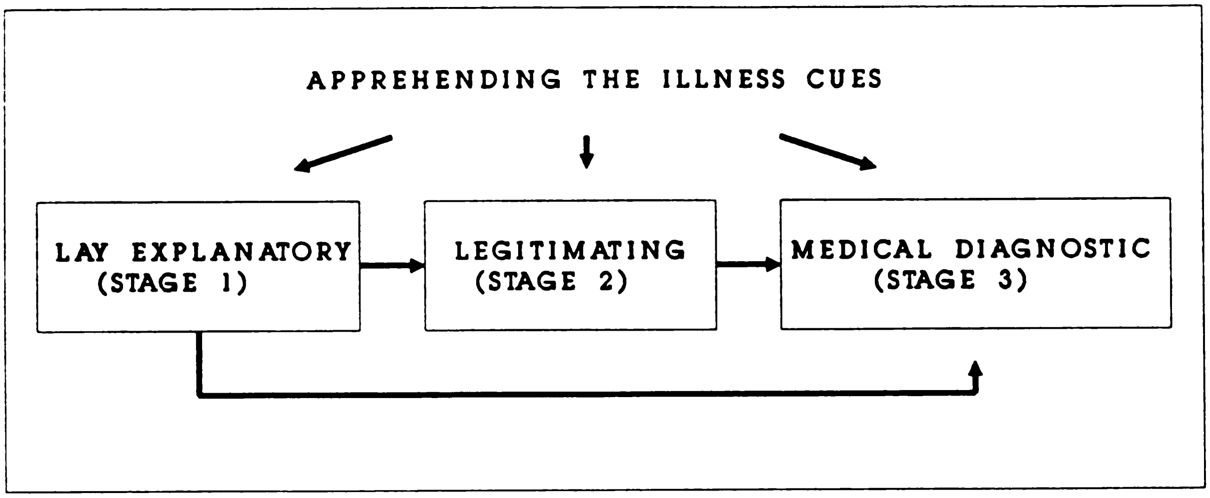


Figure 6. Possible entry points into the process of obtaining a diagnosis.

Stage 1: Lay Explanatory

The early illness cues that the child presents may be so subtle or ordinary that they do not engage the parent's attention. They simply constitute part of the background of everyday life. In most families, a cough, a bruise, a dinner barely touched, are familiar enough events that they may escape notice altogether or they may be appraised as insignificant: the result of a minor cold, a fall, a snack before mealtime. For this reason parents are often unable to describe the exact time of onset or the precise nature of the early symptoms.

The lay explanatory stage begins only after the illness cues that the child presents force their way into the conscious awareness of one of the child's caretakers, usually one of the parents, and become foreground, rather than background behaviors. The parent then focuses increasing attention on the child's behavior or symptom as something out of the ordinary and begins to ask the question, "Is something wrong with my child?" This question is first asked of the self and may subsequently be asked of others in the immediate family or social network. Lay consultations are more common when the symptoms are ambiguous.

Initially the question expresses some degree of uncertainty about the significance of what the parent is observing, but usually does not reflect a disquieting amount of concern. By asking this question, however, the parent has apprehended the potential for illness and made the transition from the pre-illness assumptive world to the lay explanatory stage of the prediagnostic period.

Once the parent acknowledges the fact that something <u>is</u> wrong, three options are available (see Figure 6). One option is for the parent to call in a physician to answer the question, "<u>What</u> is wrong with my child?". A medical diagnosis may be sought immediately, without any strategic planning for the parent-doctor interaction that is about to take place. The following conditions favor the likelihood of calling in a doctor as the first strategy after the apprehension of the illness cues: a) the parent has no stock of knowledge from which to appraise the nature of the problem, and help from a credible, trustworthy other is not available (e.g., first-time parents with no experienced family member to call upon for advice), or b) the initial appraisal of the problem is one that the parent knows he or she is not able to treat (e.g., thinks child has appendicitis), or c) the parent's characteristic tolerance for ambiguity is low or his or her need for certainty is high as a result of an actual or perceived risk of increased vulnerability, <u>and</u> d) calling a physician does not overly threaten the parents' self-esteem.

A second option is to seek medical attention, but only after considerable forethought and planning for the presentation of the child's problem in order to avoid any possible unpleasant consequences should the problem turn out to be nonexistent or minor. The unpleasant consequences include: a) being made to feel foolish or incompetent, b) the fear of not being taken seriously in the future when a "real" problem exists (the doctor will think I'm just a hypochondriac), c) losing a sense of parental competency (I'll think I'm a hypochondriac), d) feeling guilty (or being made to feel guilty) for taking up a doctor's time unnecessarily, and e) spending money unnecessarily.

One strategy that may be used to avoid these consequences is to bring up the problem behavior or the symptom at a regularly scheduled appointment and present it in a nonchalant manner, hoping that the physician will chose to pursue it further. A consequence of presenting a problem in this manner however, is that it may elicit an unconcerned response from the physician. In the case of an infant with retinoblastoma, a mother who had been concerned about her infant's failure to focus on and attend to objects for some time, reported the following interaction with the physician when she took the baby in for a routine health visit:

> When she was two months old, I took her in for her shots. I mentioned to him, "Did you notice that she was a little cross-eyed?" and he said, "Yes, most newborns are."

The third option that parents have following the apprehension of the illness cues, and the one more frequently used, is the construction of a lay explanation to answer the question of "What is wrong with my child?". This process is discussed below.

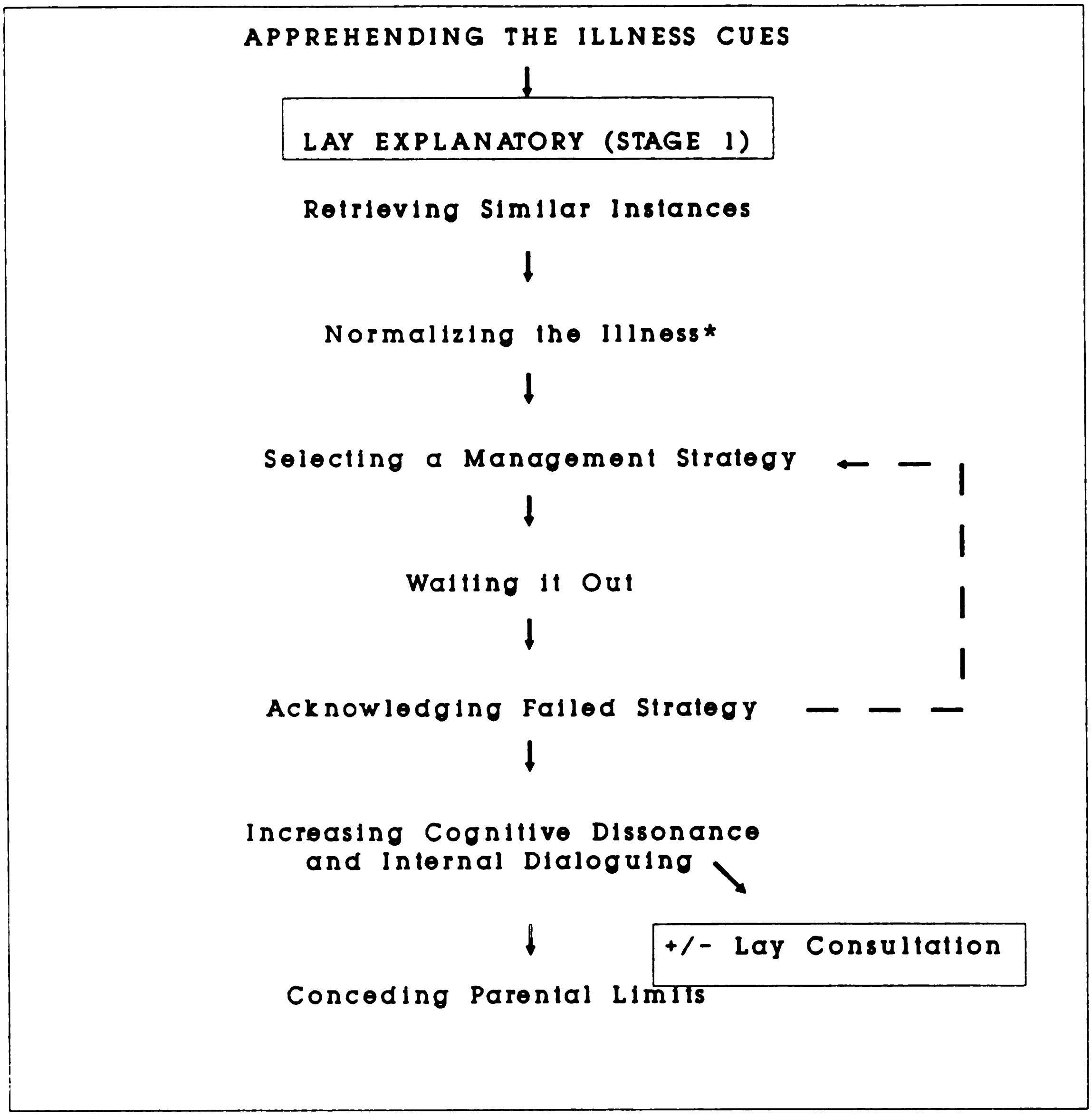
Phases of the Lay Explanatory Stage

Once the parent has apprehended the illness cues, the most common sequence or phasing of events consists of cognitively retrieving similar instances of the observed behavior or symptom, normalizing the problem or illness, selecting a management strategy, waiting it out (with or without obtaining lay consultation), acknowledging failed strategies, experiencing increasing dissonance, and internal dialoguing. The lay explanatory stage may last only a few hours, or it may continue for several days, weeks, or even months if a recursive pattern between selecting a management strategy and acknowledging the failure of that strategy occurs (see Figure 7). The transition to the next stage occurs when the parent concedes parental limits.

Retrieving Similar Instances

The cognitive retrieval of similar instances of the child's behavior or symptom involves a process of pattern recognition. In order to recognize a pattern, it must exist somewhere in the current or past biographical or sociocultural experience of the parent and be one which is fairly easily called to mind. Most parents have little or no knowledge of life-threatening childhood illnesses while knowledge of non-serious illnesses, or nuisance problems is vivid and easily retrievable. Using the availability heuristic described earlier (see p. 56), parents make potentially biased judgements about the nature of the child's problem.

In the following example a mother of an infant with cystic fibrosis who did not yet suspect that the child had a serious problem, draws upon



*If the behavior or symptom is recongized as similar to one which had serious consequences in the past, it is <u>de</u>normalized, not normalized (see Figure 8).

Figure 7. The phasing of events in the lay explanatory stage.

her stock of knowledge concerning infant crying. She cites her own behavior (or more likely what had been told to her about that behavior) as the source of her knowledge. She says of her daughter:

> . . . she was constantly crying. Constantly. And it wasn't colic or any of that stuff. It's just that she was always hungry. Well I know that some babies cry all the time and some don't, you know. I didn't and my sister did. And so, I mean I know every baby's different.

In another example, the parents of a child who was eventually diagnosed as having leukemia, initially agreed that the child's complaint of leg pains was most likely due to "growing pains". The father explained that she was "growing like mad" and the mother added that "...most mothers, when their kids have pains in their legs, think--'growing pains'." A few days later when the child became febrile, the leg pains were reappraised as a symptom of the flu. In both the initial and the subsequent interpretation of the symptom, the parents were able to recognize a pattern familiar enough to account for the child's complaint. As long as the event could be categorized as familiar, it did not pose a threat to their existing assumptive world.

Because the onset of chronic illness is usually not dramatic, parents will most often apply a familiar, minimally threatening explanation to account for the child's symptoms. They assume that what has been found to hold true in the past will adequately explain the present. The consequence of this tactic is to normalize the illness, making it comprehensible and, therefore, manageable.

Normalizing

When a symptom or a behavior is normalized, it is categorized in such a way that it fits into the parents' existing cognitive structure. Any uncertainty is thereby deflected, action is facilitated, and the incident is rendered sufficiently unproblematic to permit family life to continue with a minimal amount of disruption. Parents can account for the child's symptom or behavior by placing it within a context of what one might anticipate, or at least not be surprised by, given the current set of conditions.

By normalizing the parent does not deny that there is a problem, but categorizes it in such a way that dealing with it falls within the generally accepted scope of routine parenting. The problem may be appraised as developmental, behavioral, or medical. The parents' appraisal of the nature and severity of the problem constitutes the lay explanation. The initial lay explanations among families in this study included: abnormal eating behavior (too much or too little), abnormal amounts of crying (too much or too little), a cold, the flu, diarrhea, an allergy, teething, crossed eyes, growing pains, a reaction to an immunization, insect bites, bruises from a fall, an attention getting behavior, and a "missing muscle".

Prior to the diagnosis of leukemia, this parent explained her son's problem in behavioral terms:

> When he first started the leg pain, that was when I started working after not working for a long time, and I started thinking maybe it was an attention getting thing, the pain in his legs--or something that maybe, you know, that he was feeling like he needed more attention.

Another parent categorized her child's illness symptoms as a normal response to an immunization:

> We'd had a perfectly healthy baby. Healthy in every way. The doctor said everything was just fine and she got her DPT (immunization). That was a Friday and over the weekend she had a fever and runny stools and I just figured it was the DPT because one of our other girls had that (reaction) too.

Having diagnosed and normalized the problem, the parent begins, with varying degrees of confidence, to plan a course of action.

Selecting a Management Strategy

How parents elect to manage the problem is based on a number of conditions. If the problem is defined as medical and if the parent is knowledgeable about the usual medical therapies recommended to deal with the problem, then they are likely to use those measures. Medical strategies include such tactics as confining the child to the home and restricting activities, altering the diet, and giving over-the-counter remedies for symptomatic relief of colds, fever, and pain. For example:

> Well, she had diarrhea for three weeks. It was really pretty bad, but---I thought about bringing her to the doctor, but I thought I was being a hyperchondriac [sic], so I just waited with it. I kept giving her stuff like crackers, you know, no milk.

If, on the other hand the problem is not defined as medical, interactional strategies are more likely to be used. Interactional strategies include ignoring, providing extra attention, nagging, threatening and punishing. In her biographical account of the process leading to the diagnosis of Hodgkin's disease in one of her twin boys, Elaine Ipswitch (1979) told of the failure of both boys to make the soccer team because of being underweight. She said:

> I hoped this disappointment would motivate them to eat more. They were both picky eaters. But it did not. In fact I noticed toward the end of September that Scott was eating less than usual. His appetite stayed poor for a couple of weeks, and we began to crack down on him. At night when Scott just pushed the food around on his plate, Ronnie would tell him to stop fooling around and eat. Scott would say he couldn't. And I would say, "Yes, you can. There's no reason why you can't." Then Scott began to say he had a sore throat and it hurt to swallow. We thought this was just another one of his excuses. His throat was not red. He did not have a temperature. We just kept getting cross with him (p. 4).

Sometimes the problem is more ambiguous and the parent cannot define it as clearly medical, behavioral, or developmental. It may have components of all three categories or be so amorphous as to defy description. These are the conditions that lead parents to talk about a "gut feeling" or a "sixth sense" that something is wrong, but they cannot categorize the problem or give it a name. Under these conditions, the management strategies tend to have more of an idiosyncratic, trial and error quality.

> I knew there was a problem before we knew there was a problem . . . she had no strength. She couldn't hold herself up, you know, by the time she was eight months old . . . . First of all her heart rate was very fast. Horrible! It was even hard to hold her. She dropped off the growth charts. She was blue around the mouth. And she screamed for seven months. I knew it wasn't colic . . . . She would scream, she would close her eyes and just thrash. And there was nothing you could do. I would put her on the spin cycle on the washing machine, on the top of it, you know? I tried everything.

Waiting It Out

Once a management strategy is selected, there is a period of "waiting it out" until the problem resolves. Parents usually have a general idea of what constitutes a reasonable period of time that they must wait before expecting to see some improvement. For an infant to stop crying, the anticipated waiting period may be as short as a few minutes after he or she is picked up. In the case of an antipyretic given to reduce fever, parents may consider a reasonable waiting period to be one to two hours. For a cold or the flu to resolve, it may be a week to 10 days. Depending on the nature of the problem, it may be a shorter or longer period of time.

Acknowledging Failed Strategy

If the problem doesn't resolve or at least show improvement within the anticipated time frame, the parents must acknowledge the fact that what they thought would work, didn't. They may select another management strategy, followed by another waiting period. For the majority of parents, however, the failure of the initial strategy to relieve the symptom(s) creates a sense of dissonance between the expectation that it would, and the reality of the situation, which is that it did not. This dissonance leads to a feeling of uncertainty about their original diagnostic explanation.

Increasing Cognitive Dissonance and Internal Dialoguing

Cognitive dissonance is the recognition of a discrepancy in the relationship between two elements from what one expects that relationship should be. Festinger (1957) formally defined a dissonant relationship as one in which not-x follows from y. The dissonance expressed by the parents in this study resulted either from the failure of a specific strategy to bring about the expected consequences as it had in the past, or from a logical inconsistency between two known facts. In the following example a mother points out the dissonance that was created for her when her child, who seemed able to learn things very quickly and who had already gained bladder control, could not master bowel control.

> There wasn't any cause for any (worry) until we started to toilet train her and we couldn't figure out why she couldn't get this . . . and then when she was not able to either hold it or whatever until she was in the bathroom. When we started to toilet train her we noticed that she was able to urinate when and where she was supposed to, or when she wanted to. We knew she was a whip. We discussed photosynthesis when she was two. At first we thought it was just something that she wasn't doing because she wanted to get attention.

The inner dialogue that begins to take place as the dissonance increases was best described by Featherstone (1980).

> Two voices argue inside one head. The first sounds a warning note. Disquieted, the mother begins to contemplate some sort of action. Then a more reassuring voice answers the

> first . . . . As long as each voice retains some credibility, the dialogue continues, but time usually silences one or the other (pp.13-14).

Featherstone was speaking of developmental disorders when she referred to "time" providing the ultimate answer to the question of whether or not the child is retarded. For families whose child has a life-threatening illness, time is compressed into a much shortened period of waiting, but the internal dialoguing that she described is the same.

> Back in June we saw that Mark had a lot of bruises--on his legs especially. At first we didn't think too much of it. He crawls around in his crib and in and out of his high chair. (He's) just getting it from that. And then a couple of days later he woke up with a black eye which couldn't be accounted for, because there's no way. In his bed there's nothing sharp. That afternoon he got another black eye, the other one. But he bumped into the car. But it wasn't that hard of a bump that I thought it should get that black . . . . Leukemia came to my mind but I just completely dismissed it. I must have heard about bruises being connected with that.

When the previous explanation of the child's symptom no longer seems plausible and the inner dialogue does not produce any credible alternative possibilities, parents will concede that they have reached their explanatory limits and begin to legitimate the need for medical intervention.

Stage 2: Legitimating Medical Intervention

Having previously normalized the problem and placed its management within the scope of parental role expectations, a change must occur in the parents' appraisal of that problem before they can legitimately justify seeking medical intervention. The legitimating stage provides a transition between the lay explanatory and the medical diagnostic stages and explains the process by which the conversion takes place.

Phases of the Legitimating Stage

The legitimating stage begins when the reassuring voice of the parent's inner dialogue is silenced and the problem is denormalized. Once the problem is no longer appraised as normal, some type of strategic planning for the presentation of the child's problem to the doctor usually occurs prior to the actual parent-doctor interaction. This stage ends when the doctor is called in to determine the nature of the problem (Figure 8).

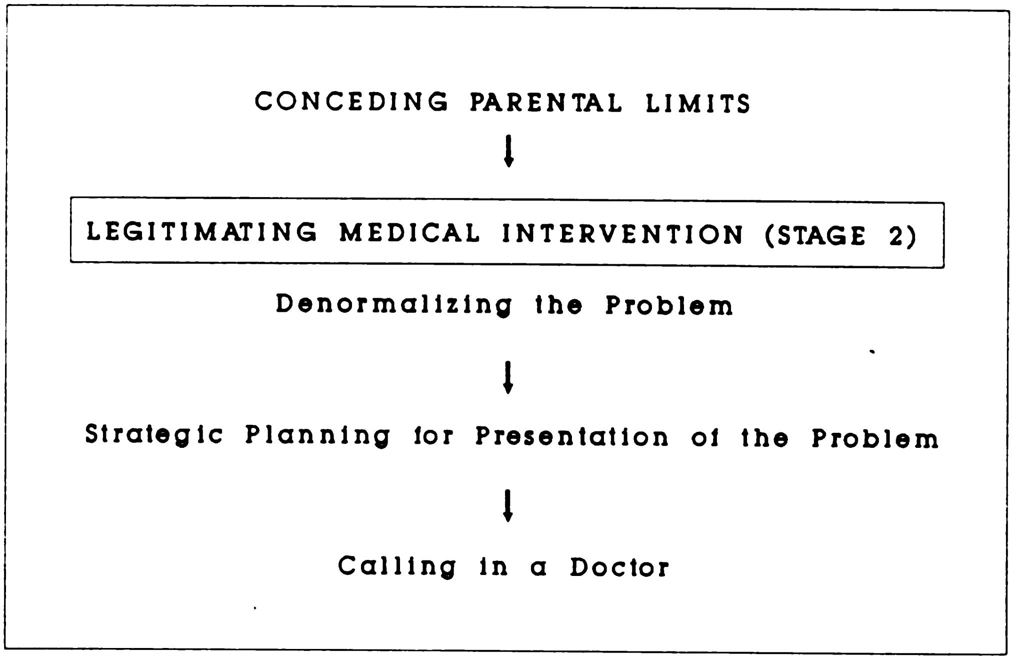


Figure 8. The phasing of events in the legitimating stage.

Denormalizing the Problem

By denormalizing the problem the parent is able to legitimate the need for professional intervention. It is no longer considered to be just a "normal" cold, or an "ordinary" bruise, or a "typical" way of gaining parental attention. If it had been, so the inner dialogue continues, it would have cleared up by now on its on, or it would have improved under the parent's management, or the symptoms would not be getting worse nor would new ones be appearing.

If the now denormalized (and therefore, medicalized) problem cannot cognitively be placed within a medical frame of reference--one that the parent has some knowledge of or experience with (e.g., appendicitis, strep throat), it will be appraised as an ambiguous threat, as indicated by statements such as, "something just doesn't seem quite right", or "something must really be wrong with him." Sometimes, however, parents have a very specific opinion about the nature of the problem. This diagnostic intuition or suspicion may exist because of either a biographical experience or a sociocultural condition that has conveyed the knowledge that bad things can and do happen to children. The consequence of either is an alteration in the parent's previous expectation or belief that "it can't or it won't happen to me," and a heightened sense of personal vulnerability.

> People will say, "Oh, chances of such and such happening are just----." To me, the chances are a lot different when something's really happened to you . . . . If somebody says one in a million, you think, well, I'm going to be that one.

A biographical reality of increased vulnerability existed for families in which there were maternal relatives known to have hemophilia. Under this condition, parents were much less likely to consider bruising or bleeding as normal.

> I have hemophilia in my family. I <u>knew</u> Tommy was a hemophiliac cause they had to give him a heel check (when he was born) . . . and he bled the whole night.

Sociocultural realities also influence the denormalizing process. The source of these realities may be information transmitted through formal or informal networks (e.g., the press, television, social conversation, etc.) or social experiences. In the following example the current AIDS epidemic has affected the parents' appraisal of the child's problem:

> Mother: So we figured he's probably allergic. We have a lot of allergies in the family, hayfever on both sides, and we thought, well maybe all these frequent colds and ear infections are just because of the allergy and . . . .
>
> Father: The other thing is we'd just started him in nursery school and (we) figured he was picking up these illnesses and infections from the other children.
>
> Mother: . . . and I thought, "My goodness, his immunity is terrible. Are there that many germs in that nursery school?" And then I thought . . . my older son went to nursery school and caught a lot of things . . . so I figured, well maybe Bobby's following that pattern.
>
> Father: I just thought it was the little kids at nursery school giving him the infections (and that) maybe he has to go through this routine till he builds up his immunity.
>
> Mother: I guess I always worried. I wondered about his immune system and I started thinking, well, you know, "What if he has some terrible disease?" But that would just flash in and out of my mind and then I would stop thinking about it.
>
> Interviewer: When you say immune system, were you thinking about AIDS?
>
> Mother: Yes. I was thinking maybe he has this--some kind of thing like that. Some kind of strange disease where his immune system isn't what it should be because he's catching all these things.

Other social experiences, such as the work environment, can alter a parent's sense of vulnerability. Working in the health care system, for example, may make a parent keenly aware of many sinister possibilities.

> Since birth, Alex had an unequal pupil . . . after (he) was born I had a dream that he had a tumor and for two months I was pretty messed up by that dream . . . . When you work in a hospital you always think, "What if this should happen to my child?"

Once the problem has been denormalized, parents then strategize how to present his or her concerns to the doctor. There are basically two reasons for strategic planning: one is to maximize the likelihood that the parent can engage the doctor's interest in investigating the problem, the other is to minimize the possibility of losing face. The parent must, therefore, convince the doctor that there is, indeed, something wrong, but must do it in a way that avoids bringing criticism upon themselves as parents. Engagement and face-saving strategies may be used alone or in combination.

Strategic Planning for Presentation of the Problem

As noted earlier, one face-saving strategy that is used with some frequency is to wait until the next routine appointment, adopt a naive stance, and bring up the problem offhandedly during the course of the visit, thereby overtly trivializing it and reducing the possibility that the parent will be judged to be an alarmist by the doctor. Another face-saving strategy is to enlist the support of credible others prior to calling in a doctor.

> We noticed unusual bruising and swelling . . . . And because she had been an ice skater and hit the ice a lot all the time, and was a very active child, bruises were not new to us, we didn't panic. But when the swelling, the unusual swelling occurred, that's when it made us stop and think. But we thought, well, we weren't going to panic. And then . . . I picked her up at school and not only did she still have a bruise, but she had an unusual goose egg, and very purple, and she said she couldn't remember having hit it or anything. And so since she is on allergy therapy we were always at the doctor's office anyway twice a week, so I thought, "Well, I'm just going to run it by the nurse, just to be careful." She looked at it (and said) "I think the doctor ought to look at it."

Engagement strategies are somewhat more complex, and require the parent to take a more knowledgeable stance. The parent must not only be prepared for the possibility that the doctor will discount his or her concerns and normalize the child's problem, but also for the possibility that he or she may not want to see the child at all. The parent must, therefore, develop a strategy that will insure that the child is examined and then manage to present counterarguments if they anticipate a normalizing appraisal by the doctor.

The following is an example of how complex and devious strategic planning can became. In this case, the parents felt that because they belonged to a prepaid medical insurance plan, there was reluctance to provide the same level of care that would have been provided on a fee for service plan.

> When I called and said, "Jeff is running a temperature of 103°, can I bring him in?" they said, "No. Keep him at home for three days and call back to (report) how he's doing." So I hung up the phone and made another call. This time I used another name. I said, "This is Mrs. Cannon. My little boy has a temperature of 102°, can I bring him in?" They asked, "What kind of insurance do you have?" When I said, "I'm going to pay cash," they said, "Bring him in."

Once the appointment is made, parents may plan interaction strategies that will decrease the likelihood that their concerns will be minimized. One way is to be prepared to list all of the diagnostic possibilities they have already considered along with the management strategies that were tried and failed. Another is for the parent to present him- or herself as having expert knowledge regarding some critical aspect of the problem. It may be the parent's intimate knowledge of the child's normal behavior (e.g., this particular child never complains or is never without energy, even when ill), that will engage the physician's interest. Or it may be the ability to present "expert" credentials. One mother, having breast fed eight children, was able to engage the physician's attention by telling him that something was not normal with the way this baby nursed. Another parent who had three brothers with hemophilia reported the following interaction following the birth of her son that was both simple and direct:

> In the hospital our doctor wanted to circumcise him and I said, "No, don't, 'cause there's a good chance that he (will have hemophilia)." And he said, "Well, I don't know what to tell you." They knew nothing at that time. So we just said, "Don't circumcise him", and that's when some of the doctors finally started saying, "Well, maybe we ought to check with someone at (the medical center)."

Whether the strategizing is simple and straightforward or complex and devious, the fact is that before calling in a doctor, usually the parents have already decided what they plan to say to legitimate the problem, and what stance they plan to assume vis-à-vis the doctor.

Stage 3: Medical Diagnostic

The medical diagnostic stage may begin in either of two ways. It may be initiated by the parent seeking medical attention for a specific concern, or by the physician, if he or she is the first to notice some abnormality. Physicians are frequently the first to detect congenital problems. If the defect is visible or easily identified by physical examination, the period of diagnostic uncertainty is usually brief and the diagnostic announcement is made to the parents shortly after birth. This is not always the case, however. If the physician has any uncertainty about the significance of his or her findings, information concerning the existence of a potential problem may be withheld from the parents until a later date when the evidence is more conclusive.

The medical diagnostic stage is the most stressful period of time for parents, not only because it is during this time that they learn, unequivocally, that their child has a life-threatening, chronic illness, but also because of the uncertainty inherent in this stage. If the period of diagnostic uncertainty is prolonged, parents often report being "driven crazy" by not knowing the nature of the problem and a sense of urgency to have a definitive diagnosis.

Phases of the Medical Diagnostic Stage

The medical diagnostic stage begins when the child is first seen by a doctor. It ends when there is diagnostic certainty and the announcement is made to the parents. The path to the correct diagnosis may be short and direct, or it may be very lengthy and involve repeated visits to the same doctor, many unsuccessful attempts to manage the problem, and several changes in health care providers, as shown in Figure 9.

Several factors are influential in determining how long this stage lasts. Among these factors are the presence or absence of unambiguous physical findings and the degree to which the physician credits or discounts the parents' concerns. The most important factor, however, seems to be the mode of medical judgment and decision making that the physician uses when presented with information about the child.

As stated earlier, the most efficacious strategy to use in diagnostic decision making employs the competing hypothesis heuristic (Wolf, Gruppen, & Billi, 1985). This heuristic requires that each piece of information be considered in relation to all possible diagnoses. Whether or not a physician uses this heuristic in the diagnostic process when the history and physical findings are ambiguous, determines the path that the process takes.

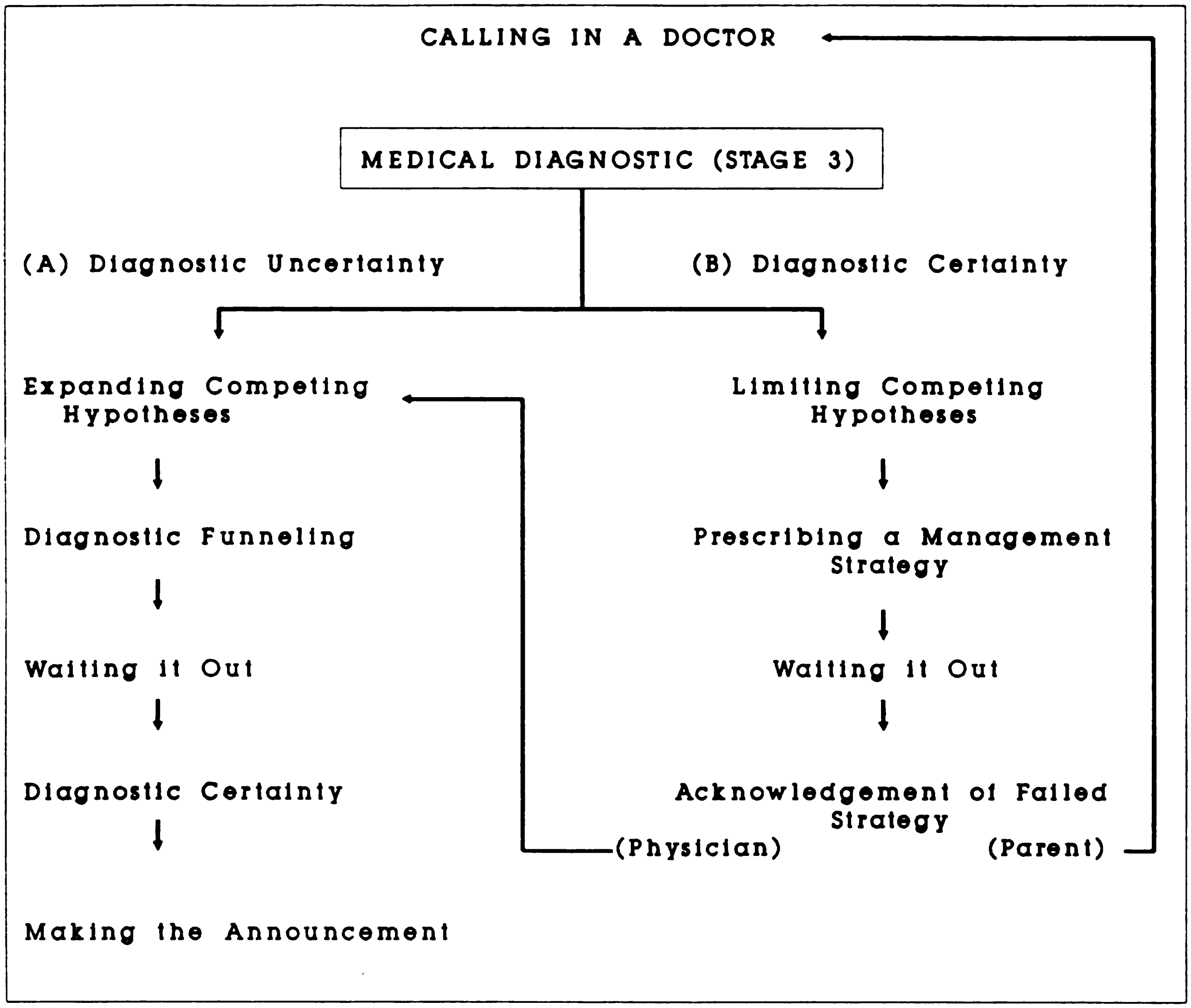


Figure 9. The phasing of events in the medical diagnostic stage. (Diagnosis is delayed if process begins at Point B.)

Limiting Competing Hypotheses (Path B)

A physician's formal knowledge that the relative frequency of a disease should not affect his or her estimate of the probability that a particular patient has the disease, is often inconsistent with his or her actual behavior. Eddy (1982) identified some of the maxims that are passed on in medical schools that contradict the rational use of probability theory: "When you hear hoof beats, think of horses, not zebras;" "Follow Sutton's law: go where the money is;" "Common things occur most commonly."

By limiting the competing hypotheses, the physician essentially uses base rates to determine the likelihood of a particular diagnosis and begins the diagnostic process with a relatively high degree of certainty about the nature of the problem. This is not an altogether unrealistic approach to the diagnosis of childhood illness when time, technical and financial resources, and physical pain and discomfort are considered. As the maxims suggest, most problems can be appropriately resolved by considering the most common causes of particular symptoms first and treating accordingly. The issue here, however, is not with the appropriateness of the physician's mode of decision making, but with the consequences for the parents when this strategy does not work because the child does, in fact, have an uncommon illness.

Of the 10 families in the Childhood Cancer study, 4 were initially told that the child had a minor problem or, in one case, no problem (i.e., a cold, constipation, an insect bite, normal newborn eye movement). Of the 21 families in the Uncertainty study, only 9 of the children were diagnosed upon the first presentation of the symptoms to a physician. Of those, 7 presented with unambiguous and severe problems at birth or within the first 3 days of life (an abdominal mass, an intestinal blockage, a ruptured ilium, hypotonia, cyanosis, persistent bleeding). The remaining 12 families had varying degrees of difficulty in resolving the uncertainty they had concerning their child's health status. The medical diagnostic period for these families ranged from a relatively brief 3 days following their initial contact with the physician, to 3 years, with many medical visits and several changes in medical providers during that time.

Prescribing a Management Strategy and Waiting It Out

The management strategy that the doctor prescribes is usually accepted by the parents, although compliance with medical strategies, such as antibiotics or dietary restrictions, is more likely than compliance with interactional ones. With both medical and interactional strategies, however, there is often a discrepant expectation between the parents and the doctor regarding how long to wait before acknowledging that the strategy has failed to achieve the expected outcome.

> We took her to the doctor on Friday and he put her on antibiotics and told her to go to bed for a week. And between Friday and Saturday, with the antibiotics, her temperature jumped from 101 to 105. On Sunday we phoned the doctor and he was out of town. We talked to another doctor and he said, "You have to give the antibiotics a chance to work. Sometimes you need more than a day or two." But, I mean, just basic education would tell you that when a child's on antibiotics and her temperature shoots up, it's not working. Either that or she's allergic to the antibiotic.

When the prescribed strategy is an interactional one, there is likely to be more disagreement as to its expected efficacy and less willingness on the part of the parent to consider using it or to continue using it for very long. For example, a parent who had been advised to ignore her two month old's crossed eyes because the problem would correct itself with time and to wait until the child was six months old before making another appointment with the physician, stated:

> So when she was three months old, you know, most babies try to reach or swing at things. She had that mobile and bright toys--she could hear, she seemed to turn, but she couldn't focus. And she was <u>really</u> cross-eyed! So I called (the doctor) back when she was three months old, and I was telling him all this and he said, "I think you're being needlessly concerned. It will straighten itself out."

Continuing to wait it out was not an acceptable strategy for this mother who then took the child to her optometrist for a second opinion.

Parental Acknowledgement of Failed Medical Strategy

Once the parents acknowledge that the medically prescribed therapy has failed, their only options are to continue to return to the same doctor or to switch the child's care to another doctor, for it is not possible for them to ignore their child's problem or to appraise it as nonthreatening any longer. As long as the parents continue to see a physician whose decision making mode is one in which competing medical hypotheses are limited, the child will remain undiagnosed and the parents will experience an ever increasing amount of uncertainty about the nature of the problem, which is now perceived as a threat, and an increasing sense of urgency to obtain an accurate diagnosis.

The following case example will serve as an illustration of an evolving set of conditions, interactions and consequences surrounding repeated failed medical strategies. Although this is a somewhat lengthy excerpt, it is presented in its entirety because contained within it are data that support the theoretical model of the prediagnostic period. This child was first taken to the doctor when she was two months old. The diagnosis of cystic fibrosis was not made until she was two and a half years old.

> When she was two months old she had a runny nose and the doctor told me it was a cold. Then when she had another cold, he said it was an allergy. She used to go the the bathroom more than regular kids and when I would tell the doctor, he said that's because I was breast feeding. When she was eight months old I told him, "I'm not breast feeding anymore, why is she still going so much?" And he told me it was because I was giving her too much liquid. So I took away the bottle and started giving her three glasses of milk a day and one glass of juice, but she was still going the same amount.

When she was a year old she was real skinny and real pale. I was going to the doctor about every three weeks. I kept telling him, "I think something's wrong with Sally." He kept saying, "No, she just has an allergy." So I'd ask him, "But why does she eat so much and go to the bathroom 5 to 10 times a day?" And he'd answer, "It's because you worry too much about her. You're making her sick." The doctor used to tell me that I was crazy. Finally, because he wouldn't listen to me, I sent my husband to see him. He told my husband that I should go back to work and not stay home with her all day. That I worried too much.

When she was two years old I asked the doctor, "Would you please run some tests for the allergy?" because she was getting worse and worse. And he said, "No, not until she's three because the tests are painful and sometimes they outgrow it." I had to sleep with her in my arms because she was coughing so much and you could hear her breathing from one room to the other. I used to tell that to the doctor, and he'd say, "Medicine isn't going to help because she's allergic to dust." I used to pay people to clean my house. I kept the house real shinny, but nothing would help.

I didn't sleep at night. Sometimes it would be three o'clock in the morning and something inside me was telling me that she was real, real sick. But I didn't know what to do because in that group there were five doctors--all those doctors, and they all kept telling me she was fine. They used to tell me that they could show me records of hundreds of kids like my daughter who don't gain weight. But I'd say, "For 18 months she hasn't grown half an inch." And they'd say, "Well, she's just going to be petite." I didn't believe them. They didn't take any tests. I told the doctor I thought she had anemia and asked him if he'd take a blood test. He said, "No, she doesn't have anemia, look at her eyes." Then I said, "Maybe she eats so much because she has parasites." He said, "No." So one time I told him I saw some worms in Sally's stool and he needed to do a test. I really didn't. I just told him that. But I needed to do something, right?

Every person that I'd see in the park or the supermarket, I'd say, "How many times does your little girl go to the bathroom? How much does she eat? Because Sally would eat 150 dollars worth of food a month. All day long she would eat, and that wasn't normal for a kid. I didn't like to ask my friends because they used to think I was crazy going to the doctor so much. But if I'd see a lady in the park, I'd feel more comfortable talking to them than talking to my friends. I thought my friends would talk about me afterwards. One lady said she was like a garbage disposal because she ate so much.

Finally we took her to another doctor. He took tests and did x-rays and just said she had a bad allergy and there was nothing he could do about it. But still something inside me

> wasn't happy. Something! I used to wake my husband up at night and say, "How can you sleep? I can't sleep. I know something is wrong with Sally."
>
> I didn't know what to do anymore. I even took her to a faith healer. Finally this lady that I met a few blocks from where I used to live, said she knew this doctor who was really good that I should go see. He was an older guy, about 85, and I took her there and I lied--I told him that I just arrived from South America and that my daughter needed a checkup because she was so skinny. He was the first one who listened to me and he knew something was wrong right away. He sent us to the medical center. And basically we didn't tell the doctors at the medical center anything different than we told all the other doctors. We told them the same stuff. The only difference is that they listened.

Physician Acknowledgement of Failed Strategy

As noted earlier, once the parents acknowledge the failed medical strategy, their only options are either to continue to try to convince their current doctor that the problem has not been adequately addressed, or to engage another physician. There are instances, however, when it is the physician who is the first to acknowledge a failed medical strategy.

Physicians, like parents, may also experience a growing sense of dissonance when what they originally appraised as a normal variant or a common or inconsequential problem, fails to resolve with a strategy of intervention or benign neglect. When this happens the doctor acknowledges his diagnostic uncertainty and expands the number of competing hypotheses that must be considered (see Figure 9). One physician, for example, initially agreed with a parent that the lump on a child's neck was probably from an insect bite and planned no intervention. However, he recorded the finding in the child's record and when she happened to be brought to the office a month later for an unrelated injury, he noted that the lump was still present and began a diagnostic workup for a malignancy.

Although physicians were not interviewed, some parents reported being aware of this increasing concern on the part of the child's doctor.

> We came home from the hospital with a normal baby and he was fine for the first few weeks. He wasn't gaining weight regularly and he had very runny stools, but the pediatrician said, "It's normal--within the range of normal. Don't worry about it, they usually lose weight right after they're born" They kept saying, "As soon as he goes off being breast fed, he'll clear up." But when he was a month old he hadn't gained quite the right amount of weight and they started observing him more closely and at about five weeks old they decided to check him into the hospital for a series of tests. They were very concerned. I wasn't at all worried. It wasn't until I was getting these vibs from them that there was this urgency to check him into the hospital to be tested that I realized something was wrong.

Expanding Competing Hypotheses (Path A)

As noted earlier, the onset of many chronic, life-threatening illnesses is gradual and initially presents with symptoms identical to common, nonserious childhood problems. For this reason, most of the children in the study enter the medical diagnostic stage via path B (see Figure 9). How long they remain in this recapitulative process is dependent upon the length of time it takes the physician to expand the medical hypotheses upon which the diagnosis is based, or upon the parents' having the good fortune to find another physician who is willing to consider an expanded list of possibilities. Until this occurs the child will remain undiagnosed and the parents will experience increasing levels of stress.

Diagnostic Funneling

Once the physician expands the diagnostic possibilities, the parents enter what Meadow (1968) has labeled the "diagnostic funnel". The child is usually referred to a medical center, subjected to multiple diagnostic procedures, and examined by a team of specialists. During this period parents may be given a number of diagnostic possibilities, along with the objective or subjective probabilities for each, or they may be given no information on the premise that it is better to wait until all the test results are in before the subject of a diagnosis is discussed.

As some medical hypotheses are discarded and the diagnostic possibilities narrow, uncertainty decreases for the physician. For the parents, however, the reverse is true. Their experience is of an expanding, diffuse, and frightening sense of uncertainty, for they have begun thinking not so much in terms of a specific diagnosis, but in terms of the meaning of a possible life-threatening illness for themselves and their child. The period of waiting for the diagnosis may become unbearable.

Waiting It Out

Robert and Suzanne Massie (1975) described most eloquently what the experience of waiting was like for them once they begin to understand the seriousness of the threat to their child's well-being:

> A day had gone by. We still knew nothing. Increasingly alarmed, we hovered over the head nurse's desk, asking anxiously, "But can't you tell us anything? What is the matter with him?" The head nurse would put on what I call now the head-nurse smirk and say, "Oh, no, there is nothing I can tell you. I think you will want to wait to hear what Doctor has to say." I particularly loathe nurses who use that expression, Doctor, without a name.
>
> So we went on waiting. Doctor did not come. In this case, Doctor was one of the most renowned hematologists of New York Hospital. Neither he nor any one of his assistants ever took a moment to call the head nurse's desk to give us a word of advice or hope. The agony of waiting by terrified parents was simply not considered. We tried to get in touch with Doctor himself; we were put off by secretaries. Over and over again the same cold phrase was repeated. "Wait" (p. 12).

Other parents faced with waiting may use a more problem-directed strategy:

> They took the test on Friday and they said we'd have to wait until Monday to reconfirm it. We were extremely upset and agitated. We didn't want to wait (even though) they said they wouldn't do anything different with him over the weekend. So a social worker helped us track down another place in the area that would do the test on Saturday.

The stress caused by prolonged waiting can become so intense that parents may experience a feeling of relief when the diagnosis becomes certain and the waiting ends. However, along with relief comes a loss of hope. Two parents expressed this paradox:

> There's a lot of difference between wondering and knowing, because when you wonder, there's always the chance that it's not (cancer). But when you know, you know (and) there's no more hope that that won't happen.
>
> I think because of the uncertainty, there was always hope. We had hope until they could finally take the CF test, the sweat test, saying, "Well maybe it's not cystic fibrosis." (But) I would still rather be well informed than be uncertain. Cause uncertainty was the killer. There you cannot have a strategy.

Diagnostic Certainty

For the physician, diagnostic certainty provides fairly clear direction for medical intervention, for each disease has a set of standard therapies or formal research protocols and the physician's actions are based on limited medical choices or computer generated decisions. For the parents, diagnostic certainty gives a name to a hitherto unknown fear, along with a statistical probability of the child's survival. It answers some of the "What" questions for the parents, but none of the "How" questions. How to manage the life of the family in the face of the multiple uncertainties that the illness brings in its wake. While diagnostic certainty brings the relief of finally knowing what was previously unknown, it also brings about the unending stress created by the need to know the unknowable.

Making the Diagnostic Announcement

There are two components to the diagnostic announcement; the informational and the interpersonal. When the illness is severe and the prognosis is poor or uncertain, it is often the parents' experience that physicians have a difficult time with one or both of these aspects of the interaction. As one parent reported:

> (The doctor) didn't quite know how to tell us what it was, and as a result, she was rather incoherent about what the disease was. My wife and I both kept asking, "What is this thing?" And she said, "Well it means if she gets colds, she's going to have more of them and it's going to be more severe." And my wife asked her, "Does this mean that she could die?" and she said, "Well she could be hit by a truck tomorrow."

Of the parents who reported receiving very accurate and detailed information, the most frequent interpersonal complaint was that the physician never looked at them during the whole interaction. Many reported that they cued in to the doctor's downcast eyes and knew that the diagnosis was ominous before a single word was spoken. John Gunther (1949) described his experience in learning that his son had a brain tumor:

> Five minutes after I got there I knew Johnny was going to die. I cannot explain this except by saying that I saw it on the faces of the three doctors, particularly Hahn's. I never met this good doctor again, but I will never forget the way he kept his face averted while he talked, and then another glimpse of his blank averted face as he said good-bye . . . (p. 29).

The diagnostic announcement, whether it is made in the context of human connection and compassion, or aloofness and tactlessness, whether information is excessive, misleading, appropriate, or withheld, the effect is the same. The announcement causes a rupture of the assumptive world and thrusts the parents into a period of chaotic activity and mind-glutting overload.

CHAPTER 6

THE RUPTURE OF THE ASSUMPTIVE WORLD

> There were no preliminaries. He announced, coldly and matter-of-factly, "The child has classical hemophilia." In one cataclysmic moment our world had been shattered. Without warning, as surely as if we had been abandoned on the bleak surface of the moon, our lives had changed. We had no idea what lay ahead.
>
> Massie & Massie, 1975

During the prediagnostic period, acknowledging the possibility that their child _might_ have a life-threatening, chronic illness posed a serious challenge to the parents' assumptive world. When the diagnostic announcement is finally made, the world as it was previously understood is suddenly and inexorably transformed. It has, in effect, ruptured.

The full impact of diagnostic certainty may be felt within a single instant or it may intrude more gradually over a period of several hours or days. Often it is experienced by parents as a physical assault by a powerful force.

> I felt as if the breath had been knocked out of me.
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> ....
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> It felt like someone had stabbed you in the heart with a knife.
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> ....
>
> I was whacked between the eyes by it.
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> ....
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> We felt like something kept knocking us to the ground.
>
> ....
>
> We had this beautiful little girl. Everything seemed to be perfect. Then all of a sudden--BAMM!

Chesler and Barbarin (1987) confirm this sense of rupture when they report that life after the diagnosis ". . . is ripped from its normal context. Parents' prior reality is shattered; they enter a new reality with new definitions of themselves and others . . . [and] they know their lives will never be what they were before."

The diagnosis becomes an assault upon previously held knowledge, beliefs, expectations, and values. It is a contradiction to the natural order and organization of family life, throwing assumed sequences and relationships out of order.

> It isn't just that children are supposed to keep on living. Imagine being eight years old and dead. It isn't just what everybody always says either--that a child dying is unnatural. It's much more than that. Old people die with achievements, memories. Children die with opportunities, dreams. They carry the hopes of all of us when they go off (Deford, 1983, p. 3).

The diagnosis defies any sense of logic or justice or fairness. The taken-for-granted world abruptly ceases to exist and the fundamental meaning upon which engagement in daily activities depends is destroyed.

> Then there was a question I asked myself incessantly. Why--of all things--should Johnny be afflicted in that part of him which was his best, the brain? What philosophical explanation could one find for that? Was all this a dismal accident, purely barren and fortuitous?. . . . But if the connection of circumstances was not fortuitous, not accidental, where was justice? (Gunther, 1949, p. 69).

Deford (1983), writing about how being the parent of a child with cystic fibrosis altered his beliefs, said:

> I think many of us have convinced ourselves that children don't die anymore, not in the latter half of the twentieth century, not in the United States of America, and certainly not in the suburbs. No never in the suburbs. But some children still do die, despite what we tell ourselves, and it makes it all the more confounding for us when an Alex confronts us with her dying . . . (pp. 20-21).

Suddenly family relationships appear to take on a temporal quality and to think about the future almost invites the threat of loss (Cohen and Wellisch, 1978). Present time becomes discontinuous with past and future time. Parents may feel a compelling need to try to control time--to stop it, slow it down, or turn it back.

Events and interactions occurring in the period immediately following the diagnostic announcement usually have an unreal quality.

Parents report living in a nightmare, acting in a play, moving through a dense fog, or feeling frozen and numb. This confusion of mind and disorganization of behavior occurs as parents "switch worlds" and move from the secure world of the known, the familiar, and the predictable to a normless world of ambiguous boundaries, unclear rules, probabilistic predictions, and sinister possibilities.

> It's like taking a blender and putting it in your brain, and all of a sudden nothing makes sense.

Diagnostic Closure and the Spread of Uncertainty

Parents and doctors experience the event of diagnostic certainty quite dissimilarly. For the physician the occasion brings closure to a series of questions and hypotheses and provides reasonably clear direction for his or her subsequent decisions and actions.

For the parents a sense of closure only occurs if the prediagnostic period had been prolonged and difficult. Under this condition the shock of the news is tempered by a feeling of relief at finally knowing for sure the nature of the child's problem and being able to institute a definite plan of treatment.

> For <u>us</u> it was a relief. I mean having her diagnosed and having her started on a course . . . a positive course of action rather than just really not knowing what was happening. I mean we didn't know if it was a neurological problem, or if she had some sort of blood disorder, or, you know, cancer.
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> . . . .
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> We didn't (have a definite diagnosis) for three months, maybe four months, because the only way you can diagnoses cystic fibrosis is with the sweat test and infants don't sweat, so they have to wait . . . until she was old enough to produce some sweat . . . . We didn't want to accept it. We were always holding out for that hope. And when they <u>did</u> finally say it <u>is</u> cystic fibrosis, we were prepared for it . . . . I think getting rid of the uncertainty--now we know what--let's deal with the cystic fibrosis now. It gave us a course of action. And my biggest problem during the whole episode was the uncertainty. Just the flat-assed uncertainty.
>
> . . . .

> All these weeks I had felt as if I were sliding down a chute in the dark, not knowing where I was or where I was going. Simply aware that there was no turning back. Now I had arrived. At a way station admittedly, not the final destination. The medical verdict could not have been worse, but in a curious way, having the diagnosis confirmed provided a respite. We were no longer in limbo (Ipswitch, 1979, p. 34).
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> . . . .
>
> Dr. Wakely told me that the reports about Sam's heart had not been exaggerated. The tests that had gone before had been inaccurate only in not showing how hideously damaged it was. I don't remember being surprised or even upset by what he said. For some strange reason it was a relief to finally know the worst (Poole, 1980, p. 66).

However the diagnostic announcement is received, whether with relief or disbelief, it creates a plethora of other uncertainties that follow in such rapid succession that the stress that had been caused by diagnostic uncertainty seems to pale by comparison. Uncertainty is no longer restricted to the nature of the illness, but spreads to every aspect of family life. Parents are now confronted with existential, etiologic, treatment, situational, biographical, and social uncertainties, but, unlike their physicians, they have no clear rules to guide their decisions and actions.

In a state of mental and emotional shock and struggling to make some sense of their new reality, parents also face the task of having to assimilate a large amount of highly specialized medical information very rapidly. This information is not only complex and unfamiliar, but much of it must, of necessity, be stated in the ambiguous language of risk and probability. This results in both an overload of information concerning what is medically known, unknown, and unknowable about the disease, and a paucity of information that can help parents make sense of what it all means within the context of their personal world. They begin the search for a new frame of reference that will give some meaning or coherence to all that is happening and grasp at any piece of information or any indicator that is appraised as having the potential to reduce the amount of perceived uncertainty.

> What I desperately needed to know then was how to live with the knowledge [emphasis added] of the disease (Massie & Massie, 1975, p. 27).
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> ....
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> We wanted to know what to expect. What we could hope for. And no one could or would tell us these things (Ipswitch, 1979, p. 25).
>
> ....
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> I went into the bathroom (at the hospital), beat the walls, used every curse word I could think of and, you know, I wanted the nurse to come...and I wanted her to show me another case just like this one. You know, I wanted to see something just like this!. . . . I didn't want to be in the same room with parents who had kids with holes in their heart. . . . I wanted something that was just as awful.
>
> ....
>
> We needed clarification to try to understand what the ground rules were going to be. We were playing a new kind of game . . . there were no parameters. We had no idea where the boundaries were and I think in discussing we were looking for where are the boundaries? Where are the sidelines? Where can't you cross over?

Existential Uncertainty

First and foremost in the mind of all parents is the question of what the disease means for the child's survival. It is a question that can only be answered by generalizations and reference to statistical probabilities, leaving the parents in limbo with regard to the long-term prognosis for their child.

> She told us that Karen had cystic fibrosis. You know the first reaction I had was--I knew it was something immense. You know, I'd heard of it but I didn't . . . I knew it was something large . . . and the first thing out of my mouth was, "How long has she got?"
>
> ....
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> We asked what were her chances of recovery and that was when it was explained that with her type of cancer, it was more like 25%, which was very discouraging.
>
> ....

> We asked (the doctor) and he said that it was really hard to predict but basically if you did nothing or do a little graft--if you did nothing he may go to about age 11 . . . . It's a last resort surgery...unfortunately he can't predict what the outcome will be.

The most common strategy to manage existential uncertainty involves transforming the probabilistic prediction (e.g., 25% chance of survival) to a relative frequency or binary outcome (either it will happen or it won't). This strategy can be best understood by considering the paradox that is created when parents are given a probability estimate for the occurrence of a unique event. Probability is defined as the frequency of an occurrence over a series of trials, but the probability of the occurrence of a unique event such as a lethal bleed, a leukemic relapse, or a surgical failure for a particular child, is either one or zero. The paradox is that while yoked to an event which has a known probability of occurrence, the child's fate is decided only once (Bursztajn, Feinbloom, Hamm & Brodsky, 1981).

> They told us that her chances were 10% to make it, which is very poor odds. But, you know, the doctor said 10% is based on groups of 100 or more people. Okay! <u>She</u> had either a 100% chance to make it or a 100% chance not to. As far as her individual case is concerned, you see, it's 50-50, more so than 10% in her individual case. She was one individual and she was either going to beat it or she wasn't.

By transforming chance to a binary form, parents limit the range of uncertainty. They now must deal with only two unambiguous possibilities: the cancer will recur or it will not, the surgery will be successful or it will not, the child will reach middle age or will not. This does not imply that a positive or negative mind set about the outcome of the illness is fixed and unchanging. On the contrary. It can change in an instant (and change back again) with or without provocation. The point is that while at any given moment in time parents are publicly conceding an 80% (or 25%, or 10%) probability of the child's survival and acknowledging that there is no way of knowing for certain what the outcome will be, they are privately operating on the assumption that the child is (or is not) going to survive and they are living their life "as if".

These private convictions may be easy to tap into, but they also may be carefully guarded and unspoken. In the latter instance, they often have a superstitious or ritualistic quality to them. For example, in response to the question, "In your own mind, do you have any thoughts about (what the outcome) is going to be?" one parent responded, "I almost feel like if I say it, well, I'll jinx it, so I don't want to say it." Another example (which is reminiscent of the ancients' strategy of devining to predict the future) is provided by Victoria Poole's (1980) account:

> I took Lynn's calendar off the wall and looked at the list I'd made of the people who'd gotten hearts since Thanksgiving.
> Burpee: Thanksgiving Day--Died Jan. 25
> Cobbie: Dec. 7--Out of ICU Feb. 2
> Terry: Jan. 7--
> Randy: Jan. 25--
> Sam: Feb. 3--
> Fred: Feb. 8--
> Died Feb. 25, I wrote after Terry's name, and then, looking at the list, I slapped my hand over my mouth. "Oh, God," I moaned. "If it's every other one, Sam isn't going to make it" (p. 289).

Another strategy used in dealing with existential uncertainty is the process of "stacking the odds." This is a strategy whereby parents are able to modify a probabilistic prognosis by gathering facts that they believe have some relevance to or bearing on the outcome of the disease. The modification may be favorable or unfavorable, it may bring relief or depression, but, at least for the moment, it serves the function of limiting the degree of uncertainty that parents must face.

> I guess girls have it worse than boys, and then we say, "Thank goodness he's a boy and not a girl." And then I guess the bad ones have it right from birth or very young and we say,"Geez, he's not a girl and he didn't have it real bad from birth." So maybe because he doesn't have it too bad, and because his (lungs) hasn't been affected too bad thus far, and because of the treatments, and the poundings, and the medicines, and the antibiotics, and the good treatment, and being close to (a medical center)...These things make us feel better.

Etiological Uncertainty

For children with genetic diseases such as hemophilia and cystic fibrosis, the etiology is clear and unequivocal. If there is also a history of the disease in other family members, etiologic uncertainty is usually not an issue for the parents. But for those in whom the disease represents a first generation expression, even a known and unambiguous pattern of inheritance may not satisfy the parents' need for an explanation of why their child was affected.

> I remember that when (the doctor) told us that in order for this to happen that both parents have to be carriers. (We) just turned and looked at each other . . . like where did we get it from? And why didn't anybody else have it? We couldn't understand....Neither of us could think of <u>anyone</u> <u>anywhere</u> down the line that had suffered from anything respiratory or anything that they died of other than my grandmother dying of cancer when she was 45. The rest of our grandparents went at a ripe old age. There are not cases of premature <u>anything</u> <u>anywhere</u>. My father, who's a scientist, who's been involved creatively with a lot of things is still convinced that CF is not genetic simply because of that.

Etiologic clarity is important not only for understanding why the disease occurred, but also for answering parents' questions concerning the future probability of the disease occurring in subsequent children or their children's children.

> Frankly we were interested in (finding out the cause of the disease) . . . because of whether we would, should have any other children . . . . That's why it took on more that academic interest at the time. We needed to know the answer to that question . . . . The possibility, the mathematical chance--it influences.

When an "acceptable" medical explanation is not possible, parents will reduce the uncertainty by creating a scenario that provides a conceivable reason for why the child was born with or developed the illness.

> I always wondered if there was a possibility that it could have been related to having had three drinks on one evening when I didn't even know I was pregnant . . . . You've got to blame it on something. I mean for lack of anything else.

Other scenarios may involve prenatal exposure to radiation, punishment for wrongdoing, a test of faith, parental age, or any other explanation that can serve as a causal link and thereby reduce etiologic uncertainty. To admit that there is no cause is to open up the possibility that one is defenseless in a chaotic and precarious world of unpredictable and uncontrollable forces.

Treatment Uncertainties

Under conditions of informational deficit, situational ambiguity, and disordered thought processes precipitated by the diagnostic announcement, parents must make significant decisions concerning treatment for their child--decisions that may be of a life and death nature and that often have a overtone of time-urgency. It is difficult to imagine a more stressful set of circumstances surrounding decision making. The following excerpt captures the dilemma:

> Well, so much happens in those first two or three days that you don't know--that's new and you're not prepared for all the decisions . . . I mean you have this social worker who comes in. You've got the oncologists that come in. You've got your clinical nurse who's there, coming in and trying to explain to you, "Do you know what a white blood cell is?" you know, I mean throwing books at you. Filling out a questionnaire, "if you wouldn't mind so that we can help." I mean in that

> first--you need a week or more to assimilate and just too much comes in at you and all you want to do is tell them, "Go away" because we're just trying to cope with what we are going through with our child right now . . . . Plus we were having to make all these decisions about randomization. But, I mean our hard thing is that--you know, it was so awful--is that we didn't know if we wanted to put him through all this (if) there was no guarantee that he was going to make it. You know, it was like...you have to deal on the very basic level of whether you even want to put him through this . . . . It was a go, no go decision . . . . There wasn't much information. You know, very little facts, very little statistics to make a real clear decision.

Making decisions about treatment entails understanding what the treatment options are, recognizing the potential benefits versus the risks that are inherent in the treatments that are available, and acknowledging the possible, but unpredictable, long-term consequences of a particular treatment choice. The difficulty in making treatment decisions is due only in part to the fact that there is so little time for the parents to absorb the amount of information that is needed in order to make an informed choice. The other difficulty is that there is much that is unknown to the medical profession and parents must base their decisions not only on information that is poorly understood, but also on medical speculation about unknown treatment effects and long-term consequences. Suzanne Massie (1975) recalls her feelings following a conversation with her pediatrician about how to manage her son's hemophilia:

> I was deeply shaken . . . . If the doctor didn't know, how could I? The doctor always knows. Every mother is sure of this. She calls the pediatrician, and he, like God, tells her when to feed and when to stop, when to add and when to take away. If the baby sneezes or wheezes, the doctor is the one who tells you, calmly and professionally, that it is nothing to worry about. If the pediatrician cannot tell you, then what? This essential rock of knowledge, this firm defense against a mother's unreasoning fear, where was it now (p. 26)?

Other dilemmas posed by treatment uncertainties are frequent sources of stress:

> It was either no treatment, the standard treatment, or the other treatment. And if we decided to randomize, we had a 50-50 chance that it would be the standard treatment anyway. So we looked at each other and asked questions relative to the pros and cons of the two treatments. And they don't know, because otherwise they'd all have one treatment.
>
> ....
>
> (He) told us that they do not have a surgery for her. Couldn't tell us much . . . just drew a picture, told us all the different things that she had and that he wanted her to come into the hospital the next day to have a band put on the pulmonary artery. And of course, at that time we couldn't make any decisions cause I hated him. You know? And all's I figured--I figured what's the point? You're telling me she's going to die. Why if she's only going to live to be 15, if we're estimating 15 at this stage of the game, why isn't it better for her to go now instead of 15?
>
> ....
>
> We were told about future problems we might have . . . side effects on the heart in future years . . . sterility . . . (problems with) her thyroid gland.
>
> ....
>
> We saw him going through all that pain the first 24 or 48 hours. We saw him going through excruciating pain and we were then being asked to make a decision about his future and to subject him to a lot more pain . . . with no guaranteed results.

When it comes to dealing with the uncertainties surrounding treatment decisions, two strategies are possible. The first is to totally relinquish any real or imagined control over the situation. This can be done by allowing "fate" to decide the course of events or by consenting to the treatment most strongly recommended by the physician. By relinquishing control to the physician or to fate, the parents' are able to reduce the stress that is created by having to make decisions under conditions of uncertainty.

> The only thing that confused me was the options that we were given. Having the baby by induced labor or going home and having the baby go as full term as possible. We were given the options of how well she might do with induced labor, but when

> talking about going full term . . . nothing else was discussed about going full term. It was like if you go full term, the baby could die . . . . They were sort of saying, you know, like they were putting it on our shoulders, which, I guess, is where it <u>should</u> have been, but it's tough to make that kind of decision . . . . But then it got to the point, I think because I was getting so big and (it was) apparent that things were getting worse for the baby, that we <u>had</u> to deliver. I mean how can you make a decision...that's a <u>Godly</u> decision as far as I'm concerned, whether someone should say whether someone should live or someone should die.
>
> ....
>
> We had complete confidence in the doctors. It's, "Here's my son. Fix him." You know, like a car. I just felt whatever (the doctor) thought should happen, should happen.
>
> ....
>
> I turned it over to God when it first happened because it was something I couldn't control....I can't cure her illness. (The doctors) know what they're doing. I don't know what I'm doing.

Although medical treatment, by its very nature, requires all parents to relinquish a certain amount of control, some do so only with great reluctance, preserving what they can at the outset and working to regain as much as they can as soon as possible. They do so by quickly becoming an expert in the control of information using tactics such as extracting, recording, reporting, censoring, expanding, limiting, and discounting. By managing the flow of information, they are able to have greater control over many aspects of their child's treatment and reduce some of the uncertainty involved in the management of the illness.

The management of information as a strategy to decrease uncertainty will be discussed in greater depth in Chapter 7. It is introduced in this section because of the difficulty that some parents experience at this early stage when, as a both a newcomer and an outsider to the medical cultural milieu, and with a sense of urgency brought about by being thrust into a cognitive void, they assertively seek information that is controlled by the professionals. The following is an extreme example, but illustrates the point well. Having just received the diagnosis, this parent recalls:

> And then we wanted to go and look up (the disease) in their library, and they locked the door and kept me out and wouldn't let me look it up in the medical books....We said <u>please</u> give us the information so we can read about this illness and the resident said, "We have to have your pediatrician authorize it." And then I was going to walk in there and just take the book out, and they locked the door.

When physicians are willing but unable to give parents adequate information on which they can base their decisions, parents who are trying to maintain some degree of control through information management may use many different information gathering strategies.

> . . . I work in a firm (that has) computerized data bases on medical conditions, and I had someone run all the materials on (her) condition, which were delivered to me because I wanted to read it all. I was very interested--we were both very interested. We were hungry for information . . . and the medical staff didn't seem to know either. Not that it was their fault. They just couldn't help us because (her condition is) so rare.

The most common reason given for wanting to gain an early and thorough access to and control of information is to protect their child. In order to protect the child, the parents must first feel certain that they have secured the very best therapy available. There is often a sense among parents that what doctors say they don't know is actually knowable if one can only tap into the right sources, for example, the latest research or the most eminent physician in the field. The second aspect of protection involves guarding against therapeutic errors and unnecessary physical and emotional trauma. By tactics such as reading relevant literature, keeping abreast of nontraditional trends, consulting with experts, learning about support services, and participating in parent groups and voluntary disease-based organizations, parents try to learn whatever they feel is necessary to control what is happening to their child.

Some parents begin to manage the informational flow early in the post diagnostic period by keeping painstakingly careful notes during the child's initial hospitalization. These notes may include everything that has been told to them, what is being done for the child, and the child's responses to treatment. The notes serve two purposes: they help the parents recall and make sense of all that has happened, and they provide leverage to insure (or demand) safe care for their child. The following example will illustrate this point:

> (We) were asking a lot of questions and we were taking down notes and we were learning as fast as we could. We were on a very quick learning curve . . . . I think it was at least three or four days before we started taking notes and that helped because then we knew what the regimen was. We knew how much medication she should have, we knew how often she should have it . . . I was just copying down everything they put on her chart so I knew her temp. We would write down whenever she got her medication. If they weren't on time I knew to go find somebody . . . . When the next round of interns came through, or the next round of residents came through, or a new nurse took over, we could tell a new intern, resident, or nurse what she was used to . . . . If there's ever any recourse or any question, that little notebook got to be <u>the</u> thing. I mean the nurses, the doctors all knew we had it and the got to where they depended on <u>us</u> many times. <u>We</u> were their source of accurate information . . . . I did the note taking for my benefit, for my peace of mind, and also as a follow up so that we knew every step of the way what was happening and if someone wasn't doing what they were supposed to be doing . . . I mean you're the only one that can watch out for your child.

Situational Uncertainty

Many children are hospitalized at the time that the diagnostic announcement is made. The unfamiliar medical environment adds the dimension of situational uncertainty to all of the uncertainties that parents are experiencing.

There are written (and unwritten) expectations that staff have concerning parental behavior, but initially parents are unaware of the rules, policies, and procedures that govern what they are expected to do, allowed to do, and prohibited from doing. They may not be at all clear about which areas they are permitted to occupy and which are off-limits. They are not sure of whom they must talk to for what kinds of questions or problems, nor do they know what their rights are as parents and as advocates for their child. They may not even know where they can find food, a bathroom, or a telephone. If they have been referred to the medical facility from some distance away, the disorientation may be even greater. The following excerpts demonstrate a small segment of the range of situational uncertainty that may confront parents:

> It was fairly late at night and there was this one young doctor up at the nurses' station and I was panicked. Something was real wrong. And I went over and I said, "Lookit. Something is going on. Not only does she have fever, but she has this and this and this . . . and he looked up and said, "I'm sorry. I can't help you. I'm on the pink team."
>
> . . . .
>
> We learned real fast how much control we had over the situation . . . you find out that you can say "no". You know, that's the biggest thing that (we) didn't know--that we had control and the ability to say "no". We didn't know how much say so we had in her treatment.

If the initial hospital stay is extended, parents may learn strategies to deal with many of the situational uncertainties during the the child's first hospitalization. For others, it comes only after repeated contact with the system.

> There are some things that are just totally unnecessary and you feel like you're pretty helpless sometimes. So you learn after a while . . . that you don't have to do <u>anything</u> there. And so if they want something done, if they want to do something unusual, you learn by experience that they have to have your approval to do it. If they're doing and IV . . . and they try it a couple of times . . . you simply stop it and tell them to get a more experienced person. So we have this rule, if they don't get in twice, they're out . . . . And you learn not to listen to interns. They think they're very knowledgeable about (the disease) and they <u>always</u> upset me . . . if I listened to all of them I wouldn't think that David would live longer than three weeks . . . . So I learned . . . to knock out that noise right away.

Parents whose child is not hospitalized when the diagnosis is made are spared having to confront the uncertainties of an unknown physical environment, but with no reference group of parents to help them adapt to their new identity, they may find their status more ambiguous than those whose children have been hospitalized. Many parents find that even though it is overwhelmingly stressful to be suddenly thrust into the world of seriously ill children, it is also helpful to be around parents who are going through the same thing. As one parent said:

> When I first walked into that (hospital) and saw all those sick babies and all that was going on, it was like walking through the gates of Hell.

This same parent also stated that being among other parents in the same situation was what helped most during the child's hospitalization:

> They understand in a way no one else can, and you don't have to explain everything.

Seeing that other parents are going through similar trials and having the same kinds of feelings helps to reduce the parents' sense of uniqueness--of having been singled out for an incomprehensible journey--and begins to establish what is frequently referred to as a "new frame of reference". This new frame of reference becomes the yardstick that parents use to gauge their progress in this passage through a seemingly normless world.

Biographical and Social Uncertainty

Biographical and social uncertainty will be addressed more fully in Chapter 7. Mention is made at this time because these dimensions of uncertainty have their origins in this immediate post diagnostic period and are always a significant source of stress. Biographical uncertainty includes, but is not limited to the feelings parents have about themselves in relation to the child's illness and their competence as parents, the concern about the effect the illness will have on the spousal relationship, and the degree of disruption that the illness will cause on future personal and family goals and plans.

> The ultimate irony (is) that simultaneously I created a life and a death, together (Deford, 1983).
>
> ....
>
> I had to get over the feeling that it was the illness that we gave him.
>
> ....
>
> It's been terrible for my husband and me. It's like we're both drowning and can't save each other. We're drifting apart. . . . We need help but we don't know where to turn.
>
> ....
>
> We had a big house in town when Eric showed up and (the doctor) asked what the circumstances were like. Well, we were proud of this brand new home we just bought and (we said), "We've got this big fancy house." And he said, "Well, that's just great. Now you can find yourselves something that's single-leveled out in the country."

Social uncertainty includes but, again, is not limited to: what kind of responses to expect from extended family, friends, and co-workers to the news of the child's illness; how to best manage those responses; how to maintain some semblance of order and normalcy for the other children during this period of upheaval; how to restructure time in order to manage the demands of work and other commitments during this period; and what the cost of treatment will be and how it will impact on the financial stability of the family. The list is without end, for with all the variability that exists among families, the diagnosis of a life threatening, chronic illness in a child can challenge the parents' biographical and social trajectories in an infinite number of ways.

Moving Beyond the Rupture

With the diagnostic announcement, the parents' assumptive world is ruptured. Rupture implies that the cognitive world <u>and</u> the action world as previously constituted no longer exist.* Time is now experienced as discontinuous rather than continuous. Parents are forced to learn a whole new stock of knowledge about parenting that is not shared by their contemporaries, thus setting them apart from others in their social world. Deeply ingrained beliefs are proven wrong, long-held values undergo major changes or are rendered inapplicable, and expectations for the future can no longer be taken for granted.

The diagnosis creates a biographical reality for these parents that transforms their assumptive world and makes it vastly different from the assumptive world of others. This results in a biographical uniqueness that sets them apart from others and creates a greater degree of divergence from the prevailing sociocultural milieu than they have ever experienced before (see Figure 10). Ipswitch (1979) described the phenomenon quite eloquently:

> We moved into another world that (others) did not share with us, could not . . . . We described it to them. They listened sensitively. But it was a world that did not take on reality until you were in it. It was as if Scott and I entered one of those old-fashioned paper-weight scenes where we could see out and others could see in, but they could not enter--and we could never completely emerge (p. 45).

---

*Although the focus of this study is on the cognitive world and how it is affected by uncertainty, it must be noted that the action world of events, behaviors, relationships, and routines is also quite suddenly shattered. Plans change or are put on an indefinite hold; significant relationships are suddenly formed out of need, while established relationships may strengthen or disintegrate; routines that were the invisible threads keeping the fabric of everyday life intact are torn apart; and the behaviors of family members may alter dramatically.

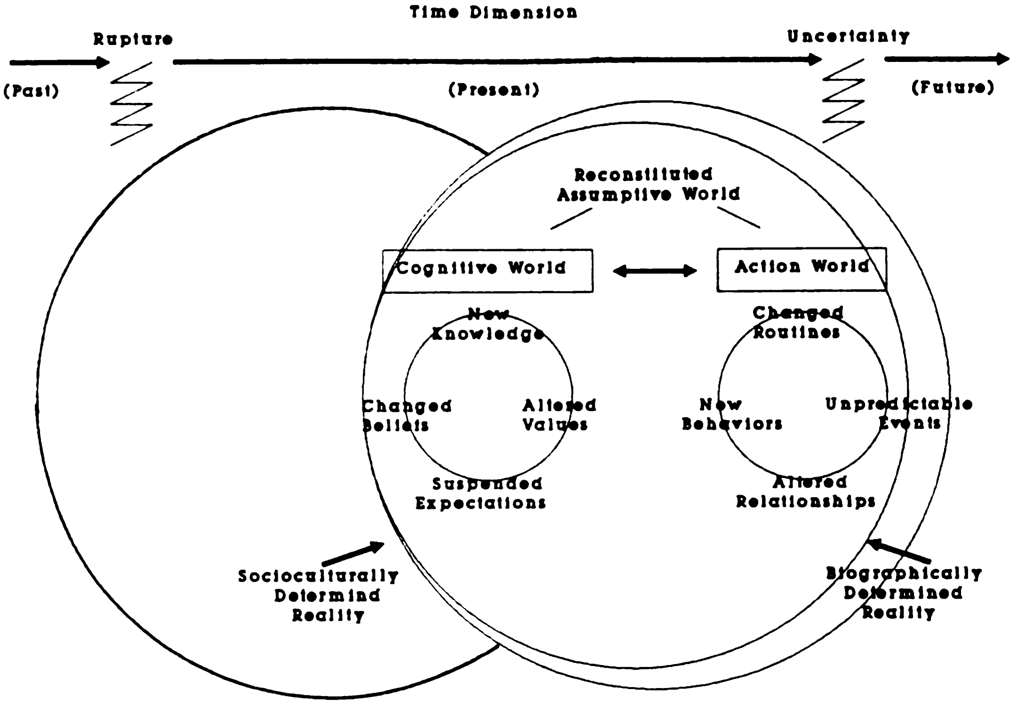

Figure 10. The Reconstituted Assumptive World showing high degree of biographical determination, marked divergence from the sociocultural norm, and discontinuity of perceived time.

The following chapter will attempt to explain how living with sustained uncertainty defines and shapes this reconstituted world.

CHAPTER 7

THE RECONSTITUTED ASSUMPTIVE WORLD:

LIVING UNDER CONDITIONS OF SUSTAINED UNCERTAINTY

> Waiting is worse than knowing. Grief rends the heart cleanly that it may begin to heal; waiting shreds the spirit.
> Llewelyn, 1978

> The only thing that makes life possible is permanent, intolerable uncertainty: not knowing what comes next.
> LeGuin, 1969

To live with a child who has a life-threatening, chronic illness is to live with sustained uncertainty. As Suzanne Massey (1975) said, "It is not the struggle but the unknown that we fear the most" (p. 245). Families have no option, however, but to learn to live with that fear, for even if the disease is under control or in remission, the knowledge that the child's status could change with little or no warning, robs parents of any respite from the reality of the threat.

> To question after question--what about the eyes? what about special therapy for the fingers? what should we particularly watch for or guard against? what if there is sudden increase in pressure? how long will it take for glioblastomatous changes to develop? when will it be safe to give more X-rays? what shall we do next?--the answers, despite the utmost good will, were confused and contradictory, simply because the course of any brain tumor in a child is unpredictable. Of course we expected too much. But it was our worst burden that we were never sure about anything, not merely from one day to another, but from one moment to the next (Gunther, 1949, p. 65).

The evidence strongly supports the conclusion that parents cannot live unchanged under such conditions of sustained uncertainty. Some of the changes come about gradually and almost imperceptibly; others occur with dramatic speed. Their reconstituted world is qualitatively different from the world that existed prior to the diagnosis during the period that Massey and Massey (1975) described as "the prehistory of our lives." The fact that not only are parents able to go on in the presence of such a sustained threat, but that the majority report that the experience has enriched their lives in many ways, is a strong commentary on the resiliency of the human spirit.

Variability in the Intensity of Perceived Uncertainty

Uncertainty is most acute and relatively constant when the disease is newly diagnosed. Koocher and O'Malley (1981) have depicted the fluctuating, but high level of stress experienced by parents at the onset of childhood cancer (see Figure 11). The level of stress caused

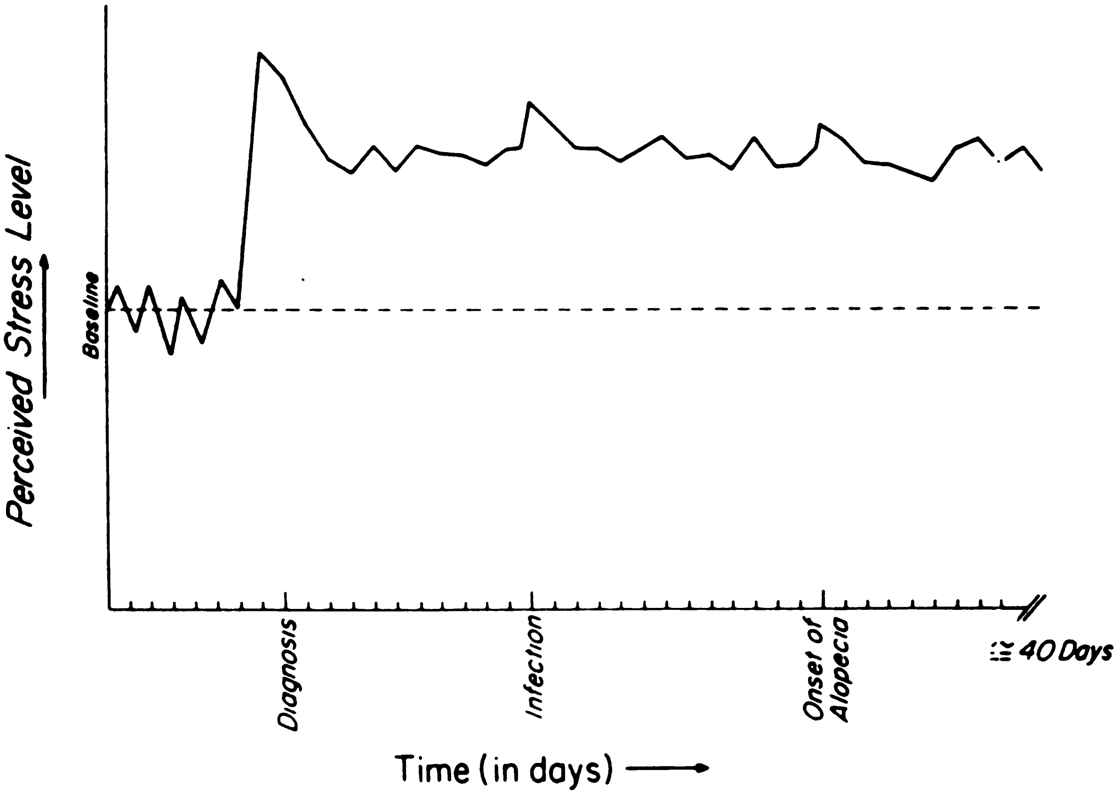

Figure 11. Hypothetical stress/time graph: Onset phase of cancer. (G. P. Koocher and J. E. O'Malley, The Damocles Syndrome, 1981, McGraw-Hill, Inc. Reprinted by permission of the publisher.)

by uncertainty can be similarly represented and, in fact, may account for most of the variability seen in Figure 11.

Uncertainty is also very intense whenever there is a change in the child's status such as a relapse, an exacerbation, an impending surgery, or the beginning of a downhill progression of the disease. The intensity of living with sustained uncertainty has been described by some parents as living with a "time bomb".

> In this illness, things can just happen overnight. He can get a lung infection and deteriorate very badly and lose a lot of weight right away, and then contract all sorts of other complications . . . . The major issue (is) we don't know which direction it will take or when. So it's a time bomb waiting to explode. And it may, or it may never explode. We just, we have no idea. So it's 100% uncertainty.
>
> ....
>
> It's like the lottery . . . that's what eats at you. I mean it's like a walking time bomb, not knowing when it's going to go off. I think that's the hardest thing of anything--not knowing.
>
> ....
>
> I think it's gonna be great news if she gets into a remission . . . . I think after <u>that</u> it's gonna be touchy. It's gonna be sitting on a powder keg and every time she gets a sniffle or sore throat or headache--I can see that as a big problem.

To talk about living with constant uncertainty is somewhat misleading, for although the <u>threat</u> posed by the disease remains constant, the <u>perception</u> of the threat does not stay at the same level of intensity. The mind simply cannot remain fixed upon any one thought for prolonged periods of time. The intrusion of other cognitive demands and distracting thoughts that occur normally in the course of a day give parents periodic respite from fear.

Once the disease has been under control or in remission for a period of time, uncertainty comes to be experienced as intruding and receding in a wave-like pattern, with distressing peaks of intensity and relatively tolerable background levels between the peaks, as depicted in

Figure 12. When the uncertainty is background, parents talk about living with the disease always "in the back of my mind." When it is rising or is at a peak, it is foreground and overshadows all other thoughts. The longer the child has been without evidence of disease, the less frequent and intense are the episodes of perceived uncertainty. However, fear lies just below the surface of consciousness and, at any time, a critical event or a memory-jogging situation can trigger renewed and intense anxiety.

> Those same fears will be there no matter what. But we try to suppress them, you know, and usually it's pretty good. And the further out you get, it's a little better. The closer you get to bone marrow day, it gets worse. After you get over that you feel like "WHEW!" I can breathe for three more months . . . . The bone marrows are once every three months and the blood tests are every month. So every time that happens it's like three little roller coasters and one big.

The Triggers of Perceived Uncertainty

There are many, commonly occurring events and interactions that can trigger a heightened level of perceived uncertainty. Among them are routine medical appointments, body variability, keywords and provocative questions, changes in therapeutic regimen, confrontation with evidence of negative outcomes, new developmental demands, and nighttime. These are discussed below, together with supporting data from parents. These data provide a predictable set of conditions that account for the variability in the intensity of perceived uncertainty.

Routine Medical Appointments

The frequency with which a child must return for follow up evaluation and care varies with the disease, its severity, and the

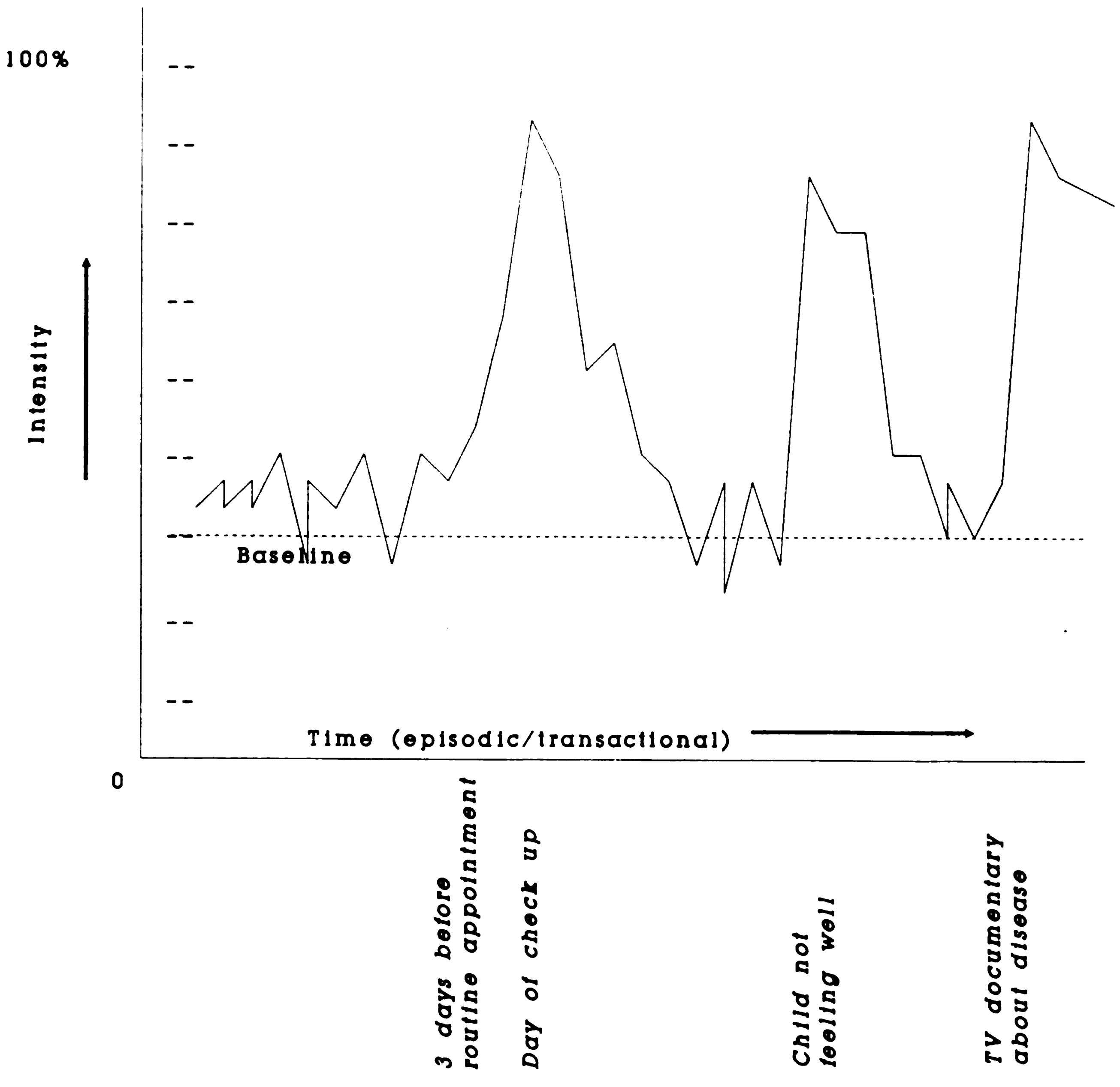

Figure 12. Hypothetical example of intensity of perceived uncertainty.

treatment regimen. However, it is accurate to say that all children must be seen at regularly scheduled intervals to be examined and have diagnostic tests performed. Anywhere from several weeks to several days prior to the scheduled appointment, worry about the outcome of the visit begins, rapidly intensifying as the day approaches. For some parents, the rise is minimal and is overshadowed by the anticipation of good news, confirming their own assessment that their child is doing well. For the majority, however, the fear reaches a peak while awaiting the results of the laboratory tests or the physical examination. The duration of the fear is extended when there is a prolonged interval between the examination and the reporting of results.

> I used to get crazy for about a week beforehand thinking about what they might find. You always worry, you always wonder, until they say "Normal" or "Normal progression."
>
> ....
>
> Those days are very difficult to get through because it brings it all back. I mean, you think, "Okay, she's been in remission, she's had checkups, and there's nothing to indicate that she isn't in remission." But, you know, it's getting that final, that final result at the end of the day . . . . Yeah, she <u>looks</u> fine--and there's nothing indicated so it <u>should</u> look fine, but there's a--but your heart skips a beat until you actually hear somebody say, "It's okay" and give you those lab counts.
>
> ....
>
> He's checked all the time (for the HIV antibody) and we die a little bit every time she tells us she's gonna do it. You know, they never call you after they've done that. If it's a <u>problem</u> she'll call us. So you wait and listen and every time that phone rings for the next two weeks, you jump right out of your skin.

<u>Body Variability</u>

Having had a child diagnosed with a serious illness affects the parents' ability to appraise any subsequent variations in the child's behavior as benign. An increase in the amount of an infant's crying, a decrease in appetite, an alteration in energy level, or the occurrence of any symptom suggestive of an illness (particularly if the symptom is similar to the one that signaled the onset of the disease), are all looked upon as a possible indicator that the disease has recurred or is progressing. Their experience has made them feel exquisitely vulnerable. Not wanting to be "caught off guard" again, they most often seek immediate medical attention rather than attempt to explain and manage the problem themselves as they had done before the rupture. They cannot return to the pre-illness world and regard the normal bumps and bruises, aches and pains, and sniffles and sneezes of childhood without alarm.

Five disease-free years after the diagnosis of cancer, one parent said, "I think we both feel like we don't worry about it--until she sneezes." Other parents report similar reactions:

> There was no such thing as a normal cold. There was nothing normal . . . there wasn't a normal fever . . . you <u>knew</u> it <u>could</u> be a normal one at some point, but you always <u>knew</u> that within a day or two it could turn bad . . . . There's just panic until someone tells you it's okay.
>
> . . . .
>
> Our whole family is more conscious of our health, not letting little aches and pains go unassessed. I mean if something's wrong, we don't wait. We go check it out. Whereas before, we kind of--"Oh well, we'll check it out later."
>
> . . . .
>
> I think he'll probably never have a quote "normal illness" because when he shrieks or there's something going on, we tense up and think it's got to be--you know--it can't just be--it <u>could</u> just be this, but it could also be lots of other things.

Keywords and Provocative Questions

The creation and reflection of reality are intimately linked to language. It is through language that knowledge, values, beliefs, and expectations are constructed, defended, modified, and reconstructed.

Parents of ill children have the harsh reality of the unpredictability or fragility of their child's status periodically reinforced by the idiosyncratic use of particular words and phrases that have become part of the medicalization of survival. Examples are terms such as "high-risk group", "long-term survivor," and "remission". This language has become so commonplace as to be unprovocative to those in the health care community; however, to many parents it implies what they already know explicitly but would rather not think about--that their child can never be considered cured and that the child's continued survival can not be assured. As Lund (1974) remarked: "I saw that each remission was harder to achieve than the last. I saw the end implicit in the word itself" (p. 191).

The use of familiar words and phrases can trigger a great deal of anxiety in parents when used in an unfamiliar context, as the following example demonstrates:

> Sometimes (the doctor) can say things that just throw me back in the seat....When she first met Alex she said, "I think we can salvage him." And, you know, now I can roll that off my tongue, very easily, but I was just shocked and Jim and I said--that night we were going, "My God! He's not a used car and everything. He's our <u>son</u>!"

Parents are extremely sensitive to hidden meanings that may lie imbedded in a communication, and a thoughtless or unfortunate choice of words can have devastating effects, as the following example demonstrates:

> Well, the only thing I wanted to do was to talk to another family that had the same thing . . . and I asked the nurse and she goes, "Well, you mean like you want to talk to someone where the kid is still alive?" So I never talked to her again . . . [and] I've never asked that again.

Very early parents come to appreciate the power that particular words or questions have in triggering anxiety in themselves and in others, and they learn to communicate with other parents in a manner that minimizes this response.

> We parents followed something of a conversational code there (at CF board meetings). Those trustees who had already lost their sick children could safely ask one another, "How's the family?" "How's everything at home?"--more or less like the lucky ones who had no CF kids. But for those of us with sick children, there was a dilemma. We did not want to charge in with "How're the kids?" because the answer might be that a child was very sick and dying. I learned, from the way some scarred old hands approached the subject with me, to offer openings that were not leading. For example, "How old are your kids now?" or, "What grade are your children in this year?" (Deford, 1983, p.44)
>
> ....
>
> This little world (the clinic waiting room community) had its own unwritten laws. You would never ask, "Does your child have leukemia?" Instead, your first question would be something like, "How long have you been coming to Children's?" And then, "Who's your doctor?" (Ipswitch, 1979, p. 49).

Changes in Therapeutic Regimen

The discontinuation of a treatment regimen known to have effected a beneficial response but no longer felt to be necessary, or the conclusion of a schedule of diagnostic tests that provided the parents with unambiguous evidence of the child's status, are both events that can trigger a great deal of diffuse anxiety. Similarly, the plan to implement a new therapy or procedure of unknown or unpredictable effectiveness can be equally alarming. At these therapeutic crossroads, the consequences of the decisions that parents make are unknowable. What parents do know is that any proposed change when their child is doing well under the current plan, threatens the precarious equilibrium that has been established. It is a paradox that so often the consequence of successfully reaching a sought after goal is an increase in anxiety about the future. For example, after the successful achievement of a four year remission from leukemia and the possibility of discontinuing a painful diagnostic procedure, this parent said:

> (The doctor) had talked about quitting having her bone marrows, but I don't want to stop that . . . . On the day that she has to have it, I'd just as soon she didn't have it, but knowing for sure that the counts come back okay and her bone marrow comes out clean---.

When their son was recategorized from the status of "inoperable" to "operable", this parent responded:

> Every time we would see (the doctor), "Don't even think about the operation." You know? He says he's not going to be a good candidate for it. But now he says he's a reasonably good candidate for it . . . . We know he'll be okay for probably two or three years or something, and seeing him doing so well---. You know he could get this operation and he might not do very well, and so, you know, he's doing real well now, so it's going to be hard for him to go into the hospital and then he's going to be sick and stuff. That's the hardest thing where he's doing real well . . . usually you wait until somebody's doing bad and then you do something. But if there's any--see they have to do it when he's in the perfect shape, because if there starts any damage, they aren't going to want to do it.

Confrontation with Evidence of Negative Outcomes

One of the conditions that allows parents (for varying periods of time) to push fear to the back of their mind is the avoidance of any evidence of the possibility of a negative outcome. Such evidence may come to the parents attention via news of the death of another child with the same diagnosis, as in the following example:

> Janice was here telling me about this girl that she worked with who was 17 and just died last year...and she'd been at the blind school, and I said, "Ohhh, how'd she lose her vision?" She said she couldn't remember . . . . Then later on we were talking and I said (the word) "retinoblastoma" and she said, "That's what Debby had!" But ohhhhh, ohhhhh--that's not what I want to hear. It's times like that when it just--sometimes you fool yourself into thinking that, well, it's not all that serious and it's gone now, and every once in a while you hear somebody say, "Yeah, so and so had that but he died about two years after that," and you go, "OOOhhhhh!"

Evidence may confront the parents through media accounts of morbidity and mortality (e.g., television feature stories, movies, documentaries, fundraising propaganda, etc.).

> Malcolm had sent us a magazine article about the wonderful successes Stanford and Shumway were experiencing, and my confidence soared. I noted, however, at the end of the paragraph, that of the 310 transplants done up to that time throughout the world, only 64 were still alive, and I was miserable all over again (Poole, 1980, p. 151).

For some parents any evidence of a possible negative outcome triggers so much distress, that they avoid exposure whenever they can.

> I try to keep my mind off it. I don't watch TV shows or anything pertaining to it. I shut them right off. For instance, my brother called and started telling me me about "Brian's Song." He'd seen it and it made him cry, and he asked if I had watched it. I said, "No." He said, "Don't you want to hear about it?," and I said, "No." So he shut up. Why hurt yourself by watching that garbage? (Chesler & Barbarin, 1987, p. 93).

Other parents may avoid such confrontations only when they are feeling particularly vulnerable, as Ipswitch (1979) describes:

> There were days when I would sit in the clinic waiting for Scott to have his tests and not be able to talk to anyone. I did not want to hear about low blood counts or children throwing up or going blind. I would hold my book in front of me and not look up, even though the print was swimming in front of my eyes. But most of the time, I looked forward to talking with the others (p. 50).

New Developmental Demands

As previously noted, once a state of relative emotional equilibrium is attained, there is often great anxiety about introducing any variation into the family's routines that could upset their potentially fragile existence. But change is inherent in the growing and developing child, and, again, it is paradoxical that the very normality that parents so desperately want for their child, also triggers increased anxiety when it occurs.

When children reach normal developmental milestones, it is necessary for parents to alter either the physical environment or their relationship with the child or both in order to promote optimum growth and development. For the parents of a child with a life-threatening illness, changes can trigger new concerns and renewed fear for the child's safety. The following excerpts provide examples of the kinds of developmental issues that can disrupt the status quo at various points in the child's development:

> We weren't really worried about him at that stage yet (when he was an infant) till he started crawling around, you know, bumping into this, bumping into that . . . . We had (his crib) padded pretty good, so I wasn't really worried about that much until he got a little older where he started to walk and crawl.
>
> ....
>
> It was real hard to let go when he first started going to school 'cause, you know, you never--I had just been with him every day. I always called and his teacher was real good about it. She would call and say, "He's doing really good and everything, don't worry." But, you know, he was still eating the high calorie breakfast before he'd go to school, and then I was always worried he wasn't drinking his milk. And I just couldn't let go.
>
> ....
>
> (He was) old enough to join the Boy Scouts, but I worried that Scott would not be well enough to keep up with the others on their hikes and camping trips . . . . The scoutmaster reassured me, saying, "Scott can participate just as much as he feels he can. There won't be any problems." But I still worried. The first overnight hike found me in a tizzy (Ipswitch, 1979, p. 54).
>
> ....
>
> I don't normally go with him (to medical appointments) anymore. . . . Jason has more control and <u>that's</u> very, very difficult for me. It's very, very, <u>very</u> difficult . . . . Doctor A. told me about three years ago, "You'll have to start to let go, Betty. He has to start to move away from you."

Nighttime

Writing about the psychological processes induced by illness, injury, and loss, Horowitz (1982) noted that, "Unbidden perceptual experiences, so common in intrusive states after trauma, occur with highest intensity and frequency when the person is relaxing his or her control, as when lying down to sleep" (p. 56). Evidence from parental data confirms that nighttime is indeed the time when parents experience some of their worst fears, however, it also suggests that it is the absence of distraction rather than the relaxation of conscious control over one's thoughts that accounts for this phenomenon. Poole (1980) states:

> The days were no problem. There was so much to do and so many people to contend with, there wasn't time to worry. By the time breakfast was over for some, it was lunchtime for others, and what was it I'd planned for dinner? . . . Having the children around me was an escape from reality, and only when I got into bed at night did the horrors come crowding in again (p. 194).

Nighttime is indeed the time of day when distractions are more likely to be absent. However, as the following example demonstrates, anytime they are absent, anxiety may increase to intolerable levels:

> If I'm real busy here with the kids, I don't think about it. Or like if I'm driving by myself, then I really think about it all the time. So I don't really try to be by myself. Like when I'm here, I'm always talking to the kids or doing something, or we're going to the park, or were running--. Driving is the worst.

The absence of distraction may be a condition that leads to a heightened perception of uncertainty, but those fears that come with night do seem to have particularly intense qualities such as "terror," "horror," and "panic."

> It's just certain nights when it all comes back to you . . . . I don't know any other word to describe it other than just terror. It's just overwhelming terror that you have. You beat it once but it's almost like it's hiding and it's waiting. It just scares you. It's just terror.
>
> . . . .
>
> It is late at night that raw panic comes. And despair. Lying in bed, sleepless, I could feel my heart pounding fiercely in my chest, dominating the stillness of the night. Waves of anxiety rolled over me. My hands were clammy. Where? What? When? When? (Massey & Massey, 1975, p. 174).

While it is important to identify those events or circumstances that are known to trigger a heightened perception of uncertainty, it is also important to recognize that many times those responses are not triggered by any identifiable set of conditions. Fear may intrude suddenly into consciousness without warning and without provocation.

> There are times where it will come up out of the clear blue sky and it will hit us--Smack!--and I think that it's very tough to come to grips right at that moment.

The Management of Uncertainty

Simply stated, uncertainty is a cognitive state brought about by the conscious awareness of not knowing the answer to a question. If the question is one that is highly salient for the survival of one's child and the future of the family, not knowing poses a threat that creates a level of uncertainty ranging anywhere from distressing to incapacitating.

That which is unknown is either knowable or unknowable. The management of uncertainty involves developing strategies to manipulate the known, the knowable, and the unknowable so that stress is reduced. This does not necessarily mean that parents want to know all that is knowable, for there are some who would prefer to live with uncertainty rather than risk knowing what they fear they will not be able to accept. Nor does it mean that what is known with certainty will be accepted without question, for often what is known may be too threatening and parents may react by creating uncertainty where none exists. Neither does the management of uncertainty imply acceptance of the realm of the unknowable as impenetrable, for parents often devise tactics to create a sense of certainty where none is possible. The management of uncertainty, therefore, refers not only to strategies that are intended to reduce uncertainty, but also to strategies that create or maintain uncertainty. Most importantly, parents may use all or any combination of these strategies under different conditions. These strategic decisions are based on the parents' appraisal of whether the consequences of knowing with certainty will empower or incapacitate them.

The management of uncertainty involves the management of six interactive dimensions of daily life. Those dimensions are time, information, awareness, social interaction, the environment, and the illness, as depicted in Figure 13. Efforts directed at managing one dimension create consequent conditions that impact on the management of others. For example, the degree to which a parent is trying to suppress thoughts about the disease (managing awareness) is likely to influence his or her decision to attend a meeting of a parent support group (managing social interaction). It is also likely that any information about possible negative outcomes will be censored (managing information). These interactions will become more apparent in the following section as each of the strategies is discussed.

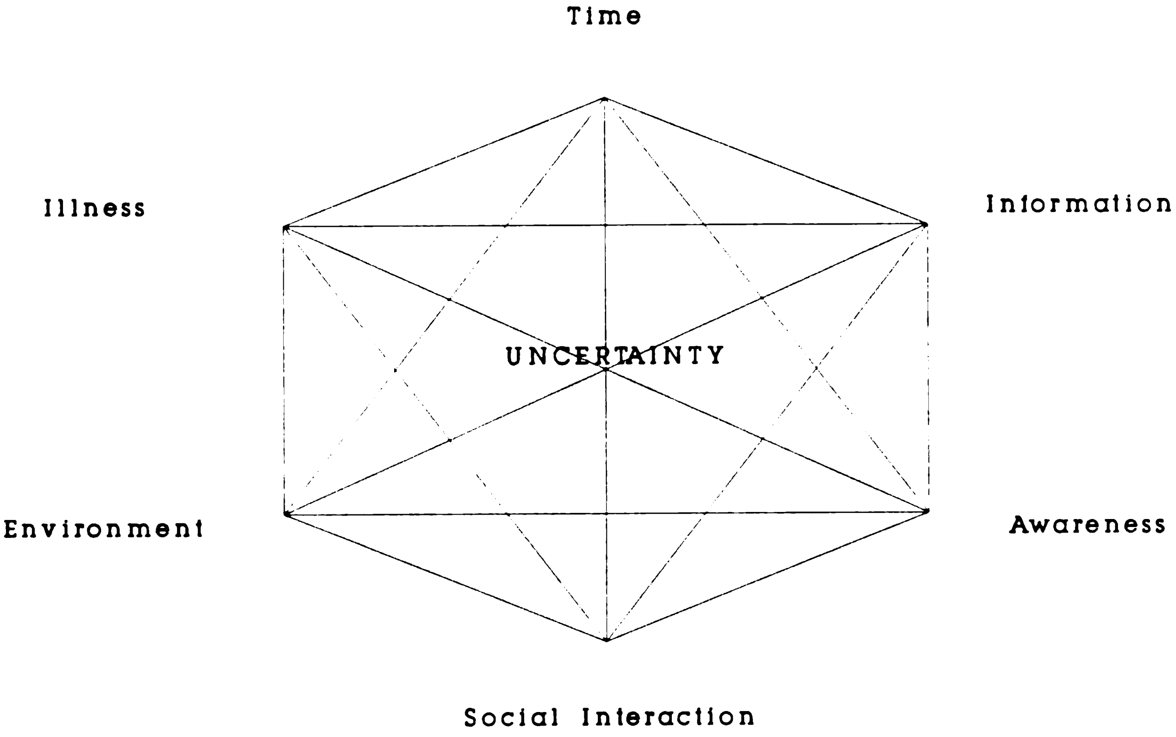

Figure 13. Strategic dimensions in the management of uncertainty.

The Management of Time

The salience of the temporal dimension of life following the rupture of the assumptive world becomes immediately explicit to parents. The span of the child's life, presumed and unquestioned before, is suddenly the main focus of parental concern. Time is now experienced as discontinuous (see Figure 10), for while the rupture prevents parents from returning to life as it was before the illness, future-oriented thinking or planing becomes too frightening given the multiple uncertainties that exist. Parents are virtually tethered to the present and the very proximate future by the rupture behind them and the uncertainties that lie ahead.

Initially the organization and practice of medicine forces the restructuring of time into shortened time units. Parents often find themselves living life in time that is measured by the intervals between laboratory tests and medical examinations. As one parent said about being in the hospital:

> Well, I get her daily blood report and it's always wonderful feedback. Everything here is by the hour, by the day. It scares me to think that getting out of the hospital, as much as I look forward to it, I'm not gonna have this daily boost. I'm sure after we get home--I guess every month, or two months, or three months, whenever the checkup is--we're gonna get pretty nervous.

Eventually parents come to realize that by adopting a "one day at a time" philosophy and living life in shortened time units, the perception of uncertainty can be reduced. They know how the child is today and tomorrow doesn't seem too far away to make a cautious prediction.

> You don't want to make big plans and put your hopes <u>too</u> far out there. You set yourself up for a big fall if you do that . . . . It's too discouraging to look at the overall picture . . . we look at him now (and say), "This is a good day. You can't tell about tomorrow. Just live right now." We take every day one day at a time. I think we have found that the daily, one day at a time philosophy has worked well for us.

The strategy of living in the day that is and restricting thoughts and plans for the future is used by all parents to some extent. When circumstances necessitate making future plans, there may be an accompanying rise in anxiety, such as Ipswitch (1979) reports:

> I began dreading any special event that Scott looked forward to--a picnic, a hike, a cookout, a party. He had to miss so many of them (p. 97).

Living in the present not only alters the parents' ability to make plans, but it has consequences for social relationships as well. Many people cannot understand the reluctance of parents to plan ahead or to commit to social events. Nor do they always understand when plans need to be suddenly changed.

> I can't make any--I don't make any plans. No plans. And the one friend I stay in touch with who doesn't have sick kids . . . just the other day I called her and invited her for dinner and I couldn't believe what I was hearing. She said, "I'm afraid to say yes because if something comes up and you can't do it, I will take it personally . . . ." And I said, "Listen, Marge, it's not you. This has been my <u>life</u>!"
>
> ....
>
> They invited us to the wedding and they wanted me to be maid of honor, but I said, "No." Cause I don't know if Jessica's going to be sick at that time. If she's sick, I'm not going to go.

The strategy of living in the present to reduce uncertainty, has another consequence for social interaction It not only effects what parents are able and willing to plan for, but it may also alter their values and beliefs about what is important in social interaction. To the extent that they find they no longer share a mutual world with their friends and extended family members, they may have difficulty interacting with them. Living in the present tends to trivialize many things. For example, denying present pleasures to save for the future may seem absurd and engaging in idle social chitchat may seem to be a wasteful use of precious time.

> I don't see the friends that I used to see . . . . I love them but I didn't want to hear about how much their root canal is going to cost.
>
> ....
>
> Hemophilia had wiped out any interest or ability I might have had for superficial relationships. It sharpened my need for knowing the essentials and made me impatient with social trivialities (Massey & Massey, 1975, p. 170).

Although the biographical future may seem too threatening to think about, the technological future, with its promise of lifesaving improvements in the management or cure of the disease, holds out the only real hope that parents have. For the majority of these families, their future is intimately linked to technology. One even expressed the feeling that there was no point in trying to imagine how the child would be ten years from now because technology could change so much in that time. Some parents expressed the feeling that they were "buying time" in the present as insurance for the future when new treatments would be available.

Managing Social Interaction

Social interaction can have a profound influence on the parents' awareness of the uncertainty surrounding their child's status. Usually they learn soon after the diagnosis that by managing certain aspects of social discourse they can reduce the number of stressful encounters. Ipswitch (1979) recalled such an encounter in a chance meeting with a doctor who belonged to her church:

> We told him about the diagnosis and how worried we were. "This is easier on us as believing Christians than on others," was his response. "Life is so short when compared to eternity." And he went on talking about how infinite eternity was. I listened in shock. He was talking as if Scott were going to die. I had been praying for Scott's recovery. I wanted to hear God would heal him, not how long eternity was (p. 16).

Managing potentially stressful social interactions involves deciding whom to tell, how much, and under what circumstances. For example, one parent said that she would not reveal that her son had hemophilia if he were just going to the movies with someone, but if he were going somewhere that might be "dangerous", she would. Disclosure decisions are often based upon the mental construction of an interactional scenario in which both positive and negative consequences are considered.

> We don't tell everybody that she has CF. We tell very close friends. <u>Very</u> close friends and family. One lady told me, "Oh, your little girl is so beautiful and she's going to die." I don't need to hear that. It's sad enough to know we have to live every day with that.
>
> ....

> We never tell (other parents) unless we have to . . . because it scares them off. You read all these articles about hemophiliacs catching AIDS (and) they're saying all hemophiliacs have AIDS. And you know, there's this couple in Florida. They burned their house down. I don't need any of that stuff.

Parents may chose to disguise or limit information rather than withhold it entirely. For example, the parents of a child with cystic fibrosis might acknowledge that the child has a bad cough, but only chose to disclose that it is not contagious. Or they may tell a teacher that the child has to take pills with meals because of a digestive problem. By deflecting the discussion away from the disease and onto a symptom, they are able to control many of the unpleasant consequences of full disclosure. Withholding, limiting, or disguising information in social situations not only lessens the risk of being caught in a potentially strained interaction or being stigmatized as a result of the disclosure, but it also protects parents from a possible anxiety-provoking confrontation with the fears they harbor for their child.

In addition to controlling when and to whom to disclose what type of information, parents must also learn to manage their own responses to the inquiries or comments of others so as not to place valued relationships in jeopardy.

> I think that because it's not obvious what's wrong with her, people misunderstand the situation and don't take it as serious as it is . . . (they say), "Oh, she looks great. She looks so precious. She's doing so good." Now I have learned to say, "Yep. Doing wonderful." I'm sure in their mind it's okay--but it's--how can I put it? It's okay for them. They don't have to think about it.

<u>The Management of Information</u>

The management of information is probably the most critical strategic dimension in the overall management of uncertainty. By controlling the quantity, quality, source, timing, and rate of flow, parents are able to establish the level or extent of knowledge that is most comfortable for them. This may range from wanting to know every known fact and every plausible theory, to wanting to know only what is absolutely essential in order to manage the illness. These extremes are contrasted in the following excerpts:

> We take her to three places--her pediatrician, the medical center, and a specialist in North Carolina--and we have all three working together and sending each other reports....And we read <u>everything</u>! I make lists of questions to ask the doctors about volumes of this or that, and what therapy we should be doing, and stuff that I picked up out of journals and magazines. Sometimes we have information on things that the doctors don't have, 'cause I call the doctors when I see articles in the press or the journals and have them explain their projects to me.
>
> ....
>
> We don't want to read. (The doctor), when we first came there, had this thick book, and he says, "It's the only one I have, and you can take it." And I said, "No thank you. I don't want it. I really don't want to get in to too much." I want to know how to deal with him, how to deal with his medication and make him feel better, and what he should do and not, but I don't want to know all these terrible things--you know, what could happen and how bad it could really-I know it's bad--but I don't really go into real detail.

It is very unusual for a parent to have the option of deciding <u>when</u> to know or <u>whether</u> to know if their child has a disease. There are two conditions, however, where this choice is possible. One is when a genetic defect can be detected before birth by prenatal testing; the other is when a screening test can be used to determine if a child, who may have been exposed to contaminated blood products, has been infected by the Human Immunodeficiency Virus. In both conditions, the parent may obtain the information if they so desire, or they may refuse the diagnostic test and wait to see if symptoms appear. By offering parents the opportunity to look into the future, these unique conditions provide the kind of situation where the issues surrounding the need to eliminate or maintain uncertainty are, perhaps, most clear. These involve the consequences of having foreknowledge of bad news. Will it empower or incapacitate? This dilemma (captured in the opening quotations of this chapter) is exemplified by a recent newspaper article that reported an interview with two mothers whose child may have been infected with HIV.

> I kept asking myself, "Can I live with the results?" And I can't . . . . Does it mean you stop kissing your child? Does it mean your child stops going to school? Does it mean your friends stop coming over?...When Joan first received the advisory letter, (that she might want to have her child tested) she called her child's doctor, who warned her: Don't test her. You'll lose your medical insurance if she's positive.
>
> Then came the news reports about Lyndon LaRouche's latest AIDS initiative on the June ballot, which could allow quarantining. "So far nuts like that have been beaten back, but what if one wins some day, and they quarantine?" Joan frets. "Does it mean they're going to take my kid away?" . . . .
>
> For Joan the uncertainty has added such stress to her life that there are days she breaks down crying . . . . Yet every time she reexamines her decision, she comes up with the same answer. "If my daughter tested negative, then my life is a lot worse now. But if she tested positive, I'd take (the stress) today over knowing that, any day."
>
> Cindy Woolley of Napa is another mother who took a different, yet also difficult route. She tested her two children, born prematurely in 1979 and 1982, but only after agonizing over the decision for nearly three years.
>
> I can't describe the soul-searching we had to do, she recalled. I kept thinking: How will it affect our family if they're positive? Will I treat them differently? . . . Then I thought: What if (researchers) come out with some treatment that could correct it? Unless I know, I couldn't do anything (San Francisco Examiner, Thursday, April 14, 1988).

Expanding Information

Parents who feel they will be empowered by information, use a variety of means to expand their knowledge. The tactics used are not unusual or unique: asking questions, reading lay and professional literature, seeking additional opinions, and networking with other parents. This behavior only becomes problematic when the information-seeking tactics of the parents conflict with the ideology of the health care provider. Physicians, nurses, and others each have their own concept of how much and what type of information is appropriate for parents to have and when it should be given. They also vary in the ease with which they are able to present this information. When parents who are seriously intent on expanding their information (and who will not be put off by responses that intimidate, deflect the question, or impugn their motive) interact with a physician or nurse who limits the information they are willing to give, they must work to find ways to extract what they want to know.

After agreeing to talk with a psychologist following a particularly traumatic event, this parent described how she was able to negotiate the information she wanted. She explained:

> (The doctor) will not discuss time with me, even if I ask. He will not discuss it at all. He makes it a firm rule. He says, "As far as I'm concerned, you take it one day at a time, and if it's 20 years, it's 20 years--and if it's 2, it's 2. And don't think about it and don't worry about it" . . . . (When the psychologists came in to talk to me) I told them, I said, "Before we talk, I want you to talk to (the doctor) and I want you to find out <u>exactly</u> how Adrian is. And then we got together in a room, and I told them, "No one leaves this room until you tell me how Adrian is, and what I can expect, and what's going on. And if (the doctor) doesn't want to tell me, let him tell you . . . I don't care <u>how</u> I get the message, but someone give me the message!"

Another frequently used tactic for extracting information is "cueing in" to the nonverbal elements of communication and to disguised messages. The most frequently cited behavior that parents cue in to as an indicator that the child is not doing well, or is not expected to do well, is a doctor's averted gaze when he or she is speaking to them. Another is a disguised message, as in the following example:

> He was sick! And the doctors opted for him to go skiing rather than go into the hospital. They were more concerned of him getting himself too tired and wearing himself out than what might happen about the infection. And, you know, that happens enough and it says to you---they're telling you something.

The discussion of how parents come to know is not complete without at least some reference to the less socially acknowledged but, nevertheless, not uncommon private experience of extracting knowledge about the future by linking it to some other known event. An example, again reminiscent of ancient methods of knowing, is provided by Poole (1980) who said:

> The jade plant seems to understand too. Both times Sam rejected, I watched horrified and disbelieving, as its leaves began to shrivel and turn yellow. When he stopped rejecting, it stopped dying . . . . It is Sam's weathervane, and, as long as its leaves are green and glossy, the future doesn't seem to be all that formidable (p. 363).

Limiting Information

Parents who fear that knowledge may incapacitate them, use tactics to limit the amount of information they are given. They don't ask many questions or read much about the illness. They may participate in a parent support group if being in the group reduces rather than creates uncertainty. For example, one parent was able to find comfort in the fact that of all the parents with children with hemophilia who attended meetings, none had a child who was HIV positive. From this she was able to conclude that the newspaper accounts of hemophiliacs being in a high risk group were greatly exaggerated.

Parents who wish to limit their knowledge of the disease, the possible complications, or the prognosis only find themselves in a problematic situation when the health care provider's beliefs about what parents should know conflict with their own. In this situation parents may be given far more information than they are able to manage.

What might have been unacceptable to another set of parents, was exactly what these parents wanted. When asked what problems they anticipated in the future, the reply was:

> They don't tell you. And I know why they don't tell you. Because if they told you, you would see those things to everybody's distraction. Also, that would be bad because you would see them and you would maybe tend to think, "Well, yes--this is what to expect," and you might put off seeing the doctor, whereas not knowing anything--and I can see where I like it better that way because if there's something that's not right, then I'm going to find out what it is. I won't attempt to diagnose it . . . or panic because I think, "Oh my gosh! This is it."

Discounting Information

There are at least three conditions that allow parents to discount negative information. One condition is that it is believed to come from an unreliable source. Inexperienced doctors and nurses and general pediatricians in private practice were often seen as not having enough experience with the child's disease to be able to give credible information. Therefore, any negative information they gave parents could be easily discounted.

A second condition that allows parents to discount negative information is having had prior experience with a failed medical prediction. One parent, for example, was told at birth that her child would probably only live for one year. At the age of eight he was doing so well that she was "convinced that Billy would be the one that would prove them wrong. That (his disease) wasn't a real disabling kind of thing, (and) that it wouldn't get worse."

The third condition for discounting information is the classification of the child as a "statistical case" or a "unique case". It is a fact that every child is unique when all the variables that impact on the disease trajectory and ultimate outcome are considered. It is also true that statistical values exist regarding morbidity and mortality rates for each of the four major diseases included in this subject population. It was apparent however, that for two of the diagnostic groups, parents were most often told the prognosis as a statistical probability (e.g., 90% of the children in her category will achieve remission), while the parents with children in the other two diagnostic groups were usually not given any statistics regarding prognosis. Instead they were told that each child was a unique case.

Parents of children who were given statistical probabilities were able to discount a poor prognosis by discounting the applicability of a statistic to a particular child.

> Percentages are percentages. They don't mean a whole lot. They're statistics, They don't <u>really</u> . . . predict anything. Because as long as it's not 100%, there's always the possibility of something else.

Parents who were told that every child was a unique case were, likewise, able to discount negative information by rationalizing that if every child was different, <u>their</u> child might be the one to prove the doctors wrong or make medical history. The following is what one parent recalls being told:

> They continually urge us to realize that every single case is completely different and unique. So that you can't infer anything from what our friends say, or from what this person says, or this piece of experience. It's a completely unique animal.

There are many paradoxes that surface in the examination of the sources and management of uncertainty in chronic childhood illness. The fact that probabilistic information, whether statistical or subjective, is both a major source of uncertainty and a means to reduce it is yet another instance.

The Management of Awareness

The management of awareness refers to all of the cognitive strategies that parents use to lessen their awareness of the threat of uncertainty. At times it involves a deliberate effort to not think about the illness or its consequences; to "push it to the back of my mind," as so many parents have said. Keeping busy and restricting attention to the tasks at hand also allows parents temporary respite from worry. Strategies may involve routinizing the activities pertaining to the child's illness so that they can be accomplished without giving them much conscious thought.

> I would say that the biggest thing is that we learn to adjust to it . . . like paying your electric bill at the end of the month. Simple as that. It just becomes a routine that you follow.

Managing awareness includes the use of beliefs that allow the parents to replace the threat of uncertainty with optimism about the future.

> I looked at it--you know when they gave you the old question in school, "Is the glass half empty or half full?" You know

> you can look at it either way. And I think very early we decided life was going to be just as full as it could be.

By managing awareness, parents are able to normalize their lives for periods of time--the time between those events which trigger a heightened perception of uncertainty and the actual crises events that occur unpredictably.

Managing the Illness

Managing the illness requires constant vigilance so that problems can be detected early and corrective action taken. Early detection is one of the major strategies to contain the disease. But early in the course of the illness, parents often do not know what they must watch for or, if they do know what to watch for, they may be unsure that they will recognize it if it does occur. So they watch for everything. Their vigil is unfocused and their anxiety diffuse.

> I watched him every minute. I didn't know what I was looking for, I just simply watched him.
>
> ....
>
> During those early months I watched Bobby constantly. I checked him every hour, all over--sometimes many times during a single hour. I woke up at night and listened for his breathing (Massey & Massey, 1975, p. 28).
>
> ....
>
> When (the doctor) said that thing (ductus arteriosus) could shut anytime, I mean you become more aware of your child. You're looking at him almost all the time, even if it's out of the corner of your eye.

With time, parents began to distinguish behaviors that might be illness related from those that are not likely to be. They learned some specific assessment skills, such as how to check a pulse rate, and began to trust that the data obtained were reliable indicators of the child's status.

Eventually their constant vigilance changes to continual scanning and periodic monitoring that is much more focused. The continual scanning is captured by the phrase, "We always have our antennae up." By having their antennae up they don't have to watch the child every minute, but <u>should</u> something occur, it is likely that it will be noticed. Monitoring is the search for a specific sign or symptom. It is quite specific and may be done at scheduled or unscheduled times.

> We give him his bath . . . and we wipe him down, cause he doesn't wipe down that good, and we notice (if) there's a bruise.
>
> ....
>
> We're better able to tell if Mark is getting ill . . . . We can check his breathing rate. So one way of knowing if he's got any involvement in his lungs is if his breathing rate starts to go up when he's resting. So we keep a pretty close monitor on that.
>
> ....
>
> If she were to get short of breath and is tired all the time. That's the sort of thing I watch for. I watch for like blueness around her mouth, I look at her fingertips once in a while . . . . I used to listen to her pulse fairly often. . . . When things don't look right, I still do.

After years of experience, parents learn to cue in to subtle variations in the child's condition. Parents talk about developing a sixth sense that enables them to know not only that something is wrong, but even before there are any visible indicators, to know that something is going to be wrong.

> As time passed, we began to realize that we could make even finer distinctions in the timing of Bobby's problems. He tended to bleed when he was emotionally upset, angry, or frustrated . . . . Finally, we realized that we (especially Sue) could tell in advance when Bobby was going to bleed. He would become pale, dark circles would shadow his eyes, and he would become cranky, irritable, and whiny (Massey & Massey, 1975, p. 112).
>
> ....
>
> (The doctor) kept saying, "Marge, it's just a throat infection. Relax, it's just a throat infection." And I kept saying, "I feel strange about this one". . . . By the next afternoon he was in lots and lots of deep trouble . . . . So

> you know. There's something inside of you--I can't explain it. There's no way you can possibly explain it. You just know!

Containing the illness also requires compliance with the treatment regimen. Early in the course of the illness, parents, fearful of the unknown consequences of varying from the prescribed schedule of treatments, adhere rigidly to medical directives. As they become more confident in their own abilities to assess the child's status, they begin to test the boundaries of therapy. They may, for example, try forbidden foods, decrease or increase a medication, eliminate a treatment, or lengthen the intervals between routine medical appointments.

Such actions, if uneventful, tend to decrease the parents' dependence on the medical center and provide them with a sense of control in managing the illness. By regaining control, parents are able to decrease the amount of perceived uncertainty. (However, it should be noted that the relationship of increased control and reduced uncertainty is not necessary an inverse relationship. Parents who completely give up control of the illness to God or Fate, for example, also seem to experience a decrease in perceived uncertainty.)

Managing the Environment

The environment poses many real and potential risks to the child with a serious health problem. When the risk is clear and the consequences certain, the uncertainty for parents involves not knowing whether they can always be present or vigilant enough to protect their child from the danger. When the risk is potential, the uncertainty arises from not knowing whether the child should be protected or, if so, to what extent. Suzanne Massey (1975) speaks of the dilemma of wanting to protect her son from environmental hazards, yet also wanting him to have a normal childhood:

> It took a constant disciplined effort to push into the far corners of my mind all the terrible ifs. Every rock, every slippery ledge, was a hazard. The uneven terrain in the forest was full of hollows and hidden holes. There were fishhooks and knives. Not to speak of the consequences of falling off a boat--the braces on Bobby's legs were like heavy anchors . . . . Try as I might, like the father of Sleeping Beauty, to hide all the spindles in the kingdom, somewhere where I least expected it a spindle lay waiting (pp. 140, 172).

Being with other children is a potential hazard to the child with a compromised immune system. Increased altitude can cause difficulty for the child with impaired cardiopulmonary function. Contracting chicken pox can be deadly to the child with leukemia. The nonavailability of a telephone may eliminate the possibility of a wilderness excursion, and the lack of a source of electricity can keep a child from a picnic. Finding a baby sitter who can care for the child in the parents' absence may be impossible. Thoughtless remarks from adults and teasing from other children are emotionally hazardous social consequences of being different.

The more tightly the parents are able to control the environment, the more certain they feel about maintaining the child's physical well-being. But the more that control interferes with normal growth and development, the more uncertain they are about the long-term consequences of their actions. Parents must make very difficult decisions regarding how much protection is necessary and how much risk is acceptable. And they must do so knowing that no one can answer those questions for them and that the consequences of a wrong decision could harm their child physically or emotionally.

The Transformed and Reconstituted World

The experience of having had a child diagnosed with a chronic, life-threatening illness, transforms the parents' assumptive world. In one way or another parents express the same sentiment: that their beliefs, values, and expectations have changed. They feel exquisitely vulnerable to misfortune, take little for granted, and feel distant from people who have not had similar experiences. However, they do feel a particular bond with those who have ill children, and they feel enriched in many ways from having been forced to confront those aspects of their lives that are truly important. The experience has made them certain that they will never be the same people that they were before.

The reconstituted world is defined by new and often unique norms. As one parent stated:

> As far as normal is concerned, if we had an outside observer, not knowing anything of what we're going through, if they walked in the door and were with us from the moment we woke up until we went to bed, they would not consider us normal at all . . . . If we had gone into somebody's house two years ago and seen this, we would say, "It's not our normal." But it's normal for us now. And that's just the way life is.

CHAPTER 8

SUMMARY AND DISCUSSION

This study began as an attempt to identify the major themes that were recurrent in the lives of families who had a child diagnosed with cancer. When the data from a 5-year longitudinal study of these families revealed that a significant long-term stressor was living under conditions of sustained uncertainty, a second study was designed to make analytically explicit the ways in which sustained uncertainty transforms the everyday life of a family. Specifically, the aim of the second study was to be able to explain the conditions that create or increase cognitive awareness of uncertainty, the social interactions that occur around issues of uncertainty, the strategies that parents use to manage the stress of uncertainty, and the consequences of living under conditions of sustained uncertainty.

In order to gain a theoretical sensitivity to the various dimensions of the concept of uncertainty once it had been identified as the central variable, a review of the literature was undertaken. This was done prior to further data collection in order to generate ideas for potential categories around which to organize the data and to suggest additional sources of data (i.e., sources to theoretically sample).

The literature review helped to identify many dimensions of the concept of uncertainty that were relevant to the study. That is to say, there was an emergent fit between the selected dimensions found in the literature and the developing theory. These dimensions included the distinction between uncertainty created by informational deficit versus that which results from situational ambiguity. Also of relevance were the reported variations among individuals to tolerate ambiguity, unpredictability, and uncontrollability.

Decision making under conditions of uncertainty identified the dimensions of risk and probability, and offered some insight into how common heuristics are used to attribute causality, form judgements, and bias decisions.

The literature also distinguished between event uncertainty and temporal uncertainty and suggested that management strategies might be differentially affected by these two properties of the concept.

The uncertainty inherent in medical practice was identified as one set of conditions for the interactions that occur between doctor and parent. Strategies frequently used to deflect medical uncertainty were described.

Following the review of the literature on uncertainty, twenty-one families who have a child with a life-threatening, chronic illness were interviewed. The analysis of data from these families uncovered a process by which parents pass from a secure, taken-for-granted, pre-illness assumptive world, through a rupture and transformation of that world, to a reconstituted, uncertain world.

The prediagnostic period begins when a parent becomes aware of a symptom or a behavior of the child that is appraised as a potential or actual indicator of a problem. For any given family, the prediagnostic period may consist of 1 to 3 stages. These stages have been designated as the lay explanatory, the legitimating, and the medical diagnostic. Although the sequence in which families pass through these stages is fixed, the time spent in any one stage is variable. A given stage may last for only a few minutes or it may continue for several years. The conditions, interactions, and strategies that are influential in determining how long a given stage lasts were identified.

The period of diagnostic uncertainty may be very brief if the lay explanatory and legitimating stages are curtailed or omitted <u>and</u> if the physician is operating under a medical ideology in which he or she expands the possible competing diagnostic hypotheses. However, diagnostic uncertainty may extend over a long period of time and be felt very intensely by parents if the physician limits the competing diagnostic hypotheses to those most frequently encountered in medical practice.

The length of the period of diagnostic uncertainty and the intensity of the stress that is created by the diagnostic delay accounts for the variability in whether or not parents feel a sense of relief when the diagnosis is made. If there was little or no diagnostic uncertainty, there is only shock and a feeling of unreality when the diagnostic announcement is made. But for those parents who have had a prolonged period of knowing that something was wrong, and perhaps <u>very</u> wrong with their child, and who have not been able to get any confirmation that their fears are grounded, the diagnostic announcement is received with a sense of relief as well as with great sadness. This finding provides partial evidence in support of the hypothesis that sustained uncertainty in chronic illness accounts for a significant amount of perceived stress.

Uncertainty may begin during pregnancy if there is reason to suspect that the fetus may have a genetic or congenital disorder. Screening tests that can resolve the uncertainty may be accepted or refused. These decisions are made on the basis of the parents' appraisal of whether the information will empower or incapacitate them. For some the certain foreknowledge of an affected child is less stressful than the continued uncertainty. For others, prenatal testing has the potential to shatter hope, making the remainder of the pregnancy unbearable. For those who feel that a loss of hope would be incapacitating, maintaining uncertainty seems preferable.

Once a child has been diagnosed as having a chronic, life-threatening illness, uncertainty becomes a dominant stressor that pervades the everyday life of the families of these children. With diagnostic closure, unidimensional uncertainty suddenly becomes multidimensional and spreads to every aspect of a family's life. The new dimensions of uncertainty that confront parents are existential, etiologic, treatment, situational, biographical, and social. The taken-for-granted cognitions and actions of the pre-illness assumptive world are challenged, ruptured, and rendered inoperable.

With their knowledge, values, beliefs, and expectations dramatically altered, parents cannot return to the way life was before the diagnosis. Nor can they take the future for granted as they previously had. They are bound to the present by the rupture behind them and the uncertainties that lie ahead.

In the early months of the illness, they can move only tentatively into the proximate future. In time, if there have not been recurrent crises, they may extend their thoughts and plans to the more distant future. Under the conditions of sustained remission or control of the disease, the frequency of periods of awareness of uncertainty lessens, but it is always just under the surface of consciousness and any critical event or memory-jogging experience can trigger renewed and intense anxiety. The most common triggers are routine medical appointments, signs of body variability, certain keywords and provocative phrases, any changes in the therapeutic regimen, confrontation with evidence of a potentially negative outcome, developmental changes in the child that demand adaptations in parenting behavior, and nighttime.

Although uncertainty is a major source of stress, there are circumstances where certainty may be appraised as more stressful. The management of uncertainty, therefore, requires strategies to reduce, create, or maintain uncertainty by manipulating the known, the unknown, and the unknowable in six interactive dimensions of daily life. These dimensions are time, information, awareness, social interaction, the environment, and the illness.

In the reconstituted assumptive world, shaped by sustained uncertainty, parents have a heightened sense of personal vulnerability which distinguishes and separates them from others who have not experienced such an event. Although changed values and altered priorities influence, and may severely strain, their relationships with others, most parents report that their experiences have brought the family closer together, strengthened their commitment to each other, and given them an appreciation for life that they find very valuable.

Implications of the Study for Nursing Practice and Research

Many of the findings of this study have implications for nursing science and nursing practice. The first is the recognition that reality is not single, nor is it fragmentable. It can not be constructed by a researcher on the assumption that parents will share a common understanding of the problem with the investigator. Rather reality is individually constructed by parents from their sociocultural milieu and their biographical history. Thus realities are multiple and holistic.

The major thrust of most nursing research concerning uncertainty has been an attempt to measure the phenomenon under varying conditions, such as hospitalization or specific diagnostic categories. This approach makes assumptions about the nature of reality that are not adequately grounded in data and ignores the most critical aspect to understanding the concept of uncertainty: that the degree of perceived stress is based upon an individual's appraisal of the situation, and that appraisal is individually constructed and holistic in nature.

This is not to say that because realities are multiple it is impossible to gain a comprehensive understanding of the ways in which uncertainty impacts on daily life. Rather it implies that the study of uncertainty must begin with the use of an appropriate paradigm that will capture, rather that control, the variance of the concept. Grounded theory provides such a paradigm and can be used to generate hypotheses that can later be tested in specific contexts.

A second finding attests to the need to recognize that the stress generated by uncertainty is a complex set of conditions that define a process rather than a unique, single event (such as diagnosis or hospitalization). This process has a biographical and sociocultural history and future. It also provides a perspective that addresses the rationality behind the fluctuating perception, duration and intensity of uncertainty and the various strategies of parents to create, maintain, or dispel uncertainty.

Since most of the sources of uncertainty cannot be eliminated, counseling regarding strategies to minimize their stressful effects should be included at appropriate points in time, and the current efforts of the family should be assessed and supported. Strategies for limiting uncertainty include the management of time, information, awareness, social interaction, the environment, and the illness.

Although the events that trigger heightened levels of uncertainty cannot be prevented from occurring within the community, exposure to some (such as prolonged periods of waiting to receive the results of a medical examination) can be predicted and controlled within the health care setting. Other events (such as developmental changes or changes in therapeutic regimen) can be anticipated well in advance of their occurrence and plans can be developed with parents regarding ways to accommodate to these changes.

The future direction of research concerning uncertainty in chronic illness should be the continued search for additional concepts and conceptual linkages in order to further develop the theory. The inclusion of additional populations, such as children with chronic illnesses that are not life-threatening and children with life-threatening, acute illnesses could broaden the theoretical base and lead to a more formal theory of uncertainty in childhood illness.

The interaction of perceived uncertainty and socioeconomic status is yet to be explored, as is the variability of uncertainty among different cultural groups. A particularly interesting area for further research that was suggested by the data, but not included in this analysis, is the mediating effects of the child on the parents' perception of uncertainty.

How families manage the stress of sustained uncertainty is likely to have long-range implications for the health and well-being of all family members and for the quality of family life. It is, therefore, a fruitful area for continued nursing research.

BIBLIOGRAPHY

Alpert, E. J. (1980). Risk and uncertainty in political choice. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger.

American Nurses' Association (1980). Nursing: A social policy statement. Kansas City, MO: Author.

Barbarin, O. A., & Chesler, M. A. (1984). Coping as interpersonal strategy: Families with childhood cancer. Family Systems Medicine, 2, 279-289.

Ben-Sira, Z. (1984). Chronic illness, stress and coping. Social Science and Medicine, 18, 725-736.

Berger, P. L., & Luckmann, T. (1966). The social construction of reality. New York: Doubleday and Co.

Beyth-Marom, R., & Dekel, S. (1985). An elementary approach to thinking under uncertainty. Hillsdale, NJ: Lawrence Erlbaum Assoc.

Borgida, E. and Nisbett, R. (1977). The differential impact of abstract vs. concrete information on decisions. Journal of Applied Social Psychology, 7, 258-271.

Boss, P., & Greenberg, J. (1984). Family boundary ambiguity: A new variable in family stress theory. Family Process, 23, 535-546.

Breznitz, S. (1983a). The seven kinds of denial. In S. Breznitz (Ed.), The denial of stress. New York: International Universities Press, Inc.

Breznitz, S. (1983b). Anticipatory stress and denial. In S. Breznitz (Ed.), The denial of stress. New York: International Universities Press, Inc.

Breznitz, S., & Goldberger, L. (1982). Stress research at a crossroads. In L. Goldberger & S. Breznitz (Eds.), Handbook of stress: Theoretical and clinical aspects. New York: The Free Press.

Buchanan, D. C., LaBarbera, C. J., Roelofs, R., & Olson, W. (1979). Reactions of families to children with Duchenne muscular dystrophy. General Hospital Psychiatry, 3, 262-269.

Budner, S. (1962). Intolerance of ambiguity as a personality variable. Journal of Personality, 30, 29-50.

Burr, C. K. (1985). Impact on the family of a chronically ill child. In N. Hobbs & J. M. Perrin (Eds.), Issues in the care of children with chronic illness. San Francisco: Jossey-Bass.

Calnan, M. (1984). Clinical uncertainty: Is it a problem in the doctor-patient relationship? Sociology of Health and Illness, 6, 74-75.

Chenitz, W. C. (1986). Getting started: The research proposal for a grounded theory study. In W. C. Chenitz & J. M. Swanson (Eds.), From practice to grounded theory. Menlo Park, CA: Addison-Wesley Publishing Co.

Chenitz, W. C., & Swanson, J. M. (1986). Qualitative research using grounded theory. In W. C. Chenitz & J. M. Swanson (Eds.), From practice to grounded theory. Menlo Park, CA: Addison-Wesley Publishing Co.

Cherlin, A. (1980). Uncertainty in family life. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger.

Chesler, M. A., & Barbarin, D. A. (1984). Difficulties of providing help in a crisis: Relationships between parents of cildren with cancer and their friends. Journal of Social Issues, 40, 113-134.

Cohen, G. (1984). Medical information and uncertainty. Medical Informatics, 9, 267-268.

Cohen, J. (1964). Behavior in uncertainty and its social implications. London: George Allen & Unwin Ltd.

Cohen, M. H., & Martinson, I. M. (1988). Chronic uncertainty: Its effect on parental appraisal of a child's health. Journal of Pediatric Nursing, 3, 89-96.

Cohen, M. L. (1983). Uncertainty rounds. Journal of the American Medical Association, 250, 1689.

Cohn, J. K., & Cohn, P. F. (1983). Patient reactions to the diagnosis of asymptomatic coronary artery disease. Implications for the primary physician and consultant cardiologist. Journal of the American College of Cardiology, 1, 956-958.

Collison, C. R. (1983). Reaching out to families for children with undiagnosed health care problems. American Journal of Maternal-Child Nursing, 8, 135-138.

Comaroff, J. & Maguire, P. (1981). Ambiguity and the search for meaning: Childhood leukaemia in the modern clinical context. Social Science and Medicine, 15, 115-123.

Connolly, T. (1980). Uncertainty, action and competence: Some alternatives to omniscience in complex problem solving. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger.

Davis, F. (1960). Uncertainty in medical prognosis: Clinical and functional. American Journal of Sociology, 66, 41-47.

Davis, F. (1963). Passage through crisis: Polio victims and their families. Indianapolis, IN: The Bobbs-Merrill Co., Inc.

Davison, S. (1984). Uncertainty (letter). Journal of the American Medical Association, 251, 2083.

Deford, F. (1983). Alex: The life of a child. New York: Viking Press.

de Vivie, E-R, & Rupprath, G. (1986). Long-term results after Fontan procedure and its modifications. Journal of Thoracic Cardiovascular Surgery, 91, 690-697.

Dewey, J. (1960). The quest for certainty. New York: Capricorn Books.

Dohrenwend, B., Pearlin, L., Clayton, P., Hamburg, B., Riley, M., & Rose, R. M. (1982). Panel report on stress and life events. In G. R. Elliot & C. Eisdorfer (Eds.), Stress and human health. New York: Springer Publishing Co.

Eddy, D. M. (1984). Variations is physician practice: The role of uncertainty. Health Affairs, 3, 74-89.

Edwards, W., Kiss, I., Majone, G., & Toda, M. (1984). A panel discussion on "what constitutes a `good decision'?" ACTA Psychologica, 56, 5-28.

Evans, P. D., Phillips, K. C., & Fearn, J. M. (1984). On choosing to make aversive events predictable or unpredictable: Some behavioral and psychophysiological findings. British Journal of Psychology, 75, 377-391.

Featherstone, H. (1981). A difference in the family. New York: Penguin Books.

Fernald, G. W., & Boat, T. F. (1987). Cystic fibrosis: Overview. Seminars in Roentgenology, 22(2), 87-96.

Ferraro, A. R., & Longo, D. C. (1985). Nursing care of the fmaily with a chronically ill, hospitalized child: An alternative approach. Image: The Journal of Nursing Scholarship, 17, 77-81.

Festinger, L. (1957). A theory of cognitive dissonance. Stanford, CA: Stanford University Press.

Fiddle, S. (1980). Introduction. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger.

Firth, M., Gardner-Medwin, D., Hosking, G., & Wilkinson, E. (1983). Interviews with parents of boys suffering from Duchenne muscular dystrophy. Developmental Medicine and Child Neurology, 25, 466-471.

Fischoff, B., Goitein, B., & Shapira, Z. (1983). Subjective expected utility: A model for decision making. In R. W. Scholz (Ed.), Decision making under uncertainty. North Holland: Elsevier Science Publishers B. V.

Forsyth, G. L., Delaney, K. D., & Greshan, M. L. (1984). Vying for a winning position: Management style of the chronically ill. Research in Nursing and Health, 7, 181-188.

Frost. C. E. B. (1980). Clinical choice under uncertainty: Its significance for regulating hospital rates. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger.

Gilgoff, I. S., & Dietrich, S. L. (1985). Neuromuscular diseases. In N. Hobbs & J. M. Perrin (Eds.), Issues in the care of children with chronic illness. San Francisco: Jossey-Bass.

Glaser, B. G. (1978). Theoretical sensitivity. Mill Valley, CA: The Sociology Press.

Glaser, B. G., & Strauss, A. L. (1967). The discovery of grounded theory. New York: Aldine Publishing Co.

Gortmaker, S. L. (1985). Demography of chronic childhood diseases. In N. Hobbs & J. M. Perrin (Eds.), Issues in the care of children with chronic illness. San Francisco: Jossey-Bass.

Gortmaker, S. L., & Sappenfield, W. (1984). Chronic childhood disorders: Prevalence and impact. The Pediatric Clinics of North America, 31, 3-18.

Guba, E. G. (1981). Criteria for assessing the trustworthiness of naturalistic inquiries. Educational Communications and Technology, 29, 75-91.

Gunther, J. (1949). Death be not proud. New York: Harper and Bros.

Haggerty, R. J. (1975). Forward. In I. B. Pless & P. Pinkerton (Eds.), Chronic childhood disorder: Promoting patterns of adjustment. Chicago: Year Book Medical Publishers.

Hansen, D. A., & Johnson, V. A. (1979). Rethinking family stress theory: Definitional aspects. In W. R. Burr, R. Hill, F. I. Nye, & I. L. Reiss (Eds.), Contemporary theories about the family (Vol. 1). New York: The Free Press.

Harrington, D. M., Block, J. H., & Block, J. (1978). Intolerance of ambiguity in preschool children: Psychometric considerations, behavioral manifestations, and parental correlates. Developmental Psychology, 14, 242-256.

Heinrichs, R. W. (1984). Verbal responses to human figure paintings: A test of the uncertainty hypothesis. Canadian Journal of Psychology, 38, 512-518.

Hobbs, N., Perrin, J. M., & Ireys, H. T. (1985). Chronically ill children and their families. San Francisco: Jossey-Bass Publishers.

Hobbs, N., Perrin, J. M., Ireys, H. T., Moynihan, L. C., & Shayne, M. W. (1984). Chronically ill children in America. Rehabilitation Literature, 45, 206-213.

Hoffman, I., & Futterman, E. H. (1971). Coping with waiting: Psychiatric intervention and study in the waiting room of a pediatric oncology clinic. Comprehensive Psychiatry, 12, 67-81.

Huang, N. N., Schidlow, D. V., Szatrowski, T. H., Palmer, J., Laraya-Cuasay, L. R., Yeung, W., Hardy, K., Quitell, L., & Fiel, S. (1987). Clinical features, survival rate, and prognostic factors in young adults with cystic fibrosis. The American Journal of Medicine, 82, 871-877.

Humes, R. A., Porter, C. J., Mair, D. D., Rice. J. J., Offord, K. P., Puga, F. J., Schaff, H. V., & Danielson, G. K. (1987). Intermediate follow-up and predicted survival after the modified Fontan procedure for tricuspid atresia and double-inlet ventricle. Circulation, 76(Suppl. III), III-67-III-71.

Ipswitch, E. (1979). Scott was here. New York: Delacorte Press.

Jacox, A. (1981, June). Competing theories of science. Paper presented at 1981 Forum on Doctoral Education in Nursing, Seattle, WA.

Jahoda, M. (1980). Some comments on the role of social psychology on forecasting. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger.

Janis, I. L. (1982). Decision making under stress. In L. Goldberger & S. Breznitz (Eds.), Handbook of stress: Theoretical and clinical aspects. New York: Free Press.

Jessop, D. J., & Stein, R. E. K. (1985). Uncertainty and its relation to the psychological and social correlates of chronic illness in children. Social Science and Medicine, 20, 993-999.

Jungermann, H. (1983). The two camps on rationality. In R. W. Scholz (Ed.), Decision making under uncertainty. North Holland: Elsevier Science Publishers B. V.

Katz, J. (1984). Why doctors don't disclose uncertainty. The Hastings Center Report, 14, 35-44.

Kiev, A. (1976). Cultural perspectives on the range of human behavior. Mental Health and Society, 3, 53-56.

Klayman, J. (1984). Learning from feedback in probabalistic environments. ACTA Psychologica, 56, 81-92.

Klenow, D. J. & Dasilva, F. B. (1980). Uncertain timetables: A case of spare part surgery. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger Publishers.

Knox, J. E., & Hayes, V. E. (1983). Hospitalization of a chronically ill child: A stressful time for parents. Issues in Comprehensive Pediatric Nursing, 6, 217-226.

Koocher, G. P. (1985). Psychosocial care of the child cured of cancer. Pediatric Nursing, 11, 91-93.

Koocher, G. P., & O'Malley, J. E. (1981). The Damocles syndrome: Psychosocial consequences of surviving childhood cancer. New York: McGraw-Hill Book Co.

Lau, R. R., Williams, S., Williams, L. C., Ware, J. E., & Brook, R. H. (1982). Psychosocial problems of chronically ill children: Physician concern, parent satisfaction, and the validity of medical records. Journal of Community Health, 7, 250-261.

Lazarus, R. S., and Folkman, S. (1984). Stress, appraisal, and coping. New York: Springer Publishing Co.

Lerner, A. (1980). Orientation to ambiguity. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger Publishers.

Light, D., Jr. (1979). Uncertainty and control in professional training. Journal of Health and Social Behavior, 20, 310-322.

Lincoln, Y. S., & Guba, E. G. (1985). Naturalistic inquiry. Beverly Hills, CA: Sage Publications.

Lippman-Hand, A., & Fraser, F. C. (1979a). Genetic counseling--the postcounseling period: I. Parent's perceptions of uncertainty. American Journal of Medical Genetics, 4, 51-71.

Lippman-Hand, A., & Fraser, F. C. (1979b). Genetic counseling--the postcounseling period: II. Making reproductive choices. American Journal of Medical Genetics, 4, 73-87.

Llywelyn, M. (1978). The wind from Hastings. Boston: Houghton Mifflin.

Longhofer, J. (1980). Dying or living?: The double bind. Culture, Medicine, and Psychiatry, 4, 119-136.

Lorenzi, P. (1980). Applied behavior under uncertainty. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger Publishers.

Lund, D. H. (1974). Eric. Philadelphia: Lippincott Co.

Madden, N. A., Terrizzi, J., & Friedman, S. B. (1982). Psychological issues in mothers of children with hemophilia. Journal of Developmental and Behavioral Pediatrics, 3, 136-142.

Mages N. L., & Mendelsohn, G. A. (1979). Effects of cancer of patient's lives: A personological approach. In G. C. Stone, F. Cohen, & N. E. Adler (Eds.), Health psychology: A handbook. San Francisco: Jossey-Bass.

Martinson, I. M., & Cohen, M. H. (1988). Themes from a longitudinal study of family reaction to childhood cancer. Journal of Psychosocial Oncology. 3 (in press).

Martinson, I., Gilliss, C. L., Moore, I., Cohen, M., Coughlin, D., & Rudin, M. (1985). Impact of childhood cancer on American families (Contract No. 2-210-PR-14). San Francisco: California Division of American Cancer Society.

Mason, C. (1985). The production and effects of uncertainty with special reference to diabetes mellitus. Social Science and Medicine, 21, 1329-1334.

Massie, R., & Massie, S. (1975). Journey. New York: Alfred A. Knopf.

Mastromauro, C., Myers, R. H., & Berkman, B. (1986). Attitudes toward presymptomatic testing in Huntington disease. American Journal of Medical Genetics, 26, 271-282.

Mattsson, A. (1972). Long-term illness in childhood: A challenge to psychosocial adaptation. Pediatrics, 50, 801-811.

McAnarney, E. R., Pless, I. B., Satterwhite, B., & Friedman, S. B. (1974). Psychological problems of children with chronic juvenile arthritis. Pediatrics, 53, 523-528.

McKeever, P. T. (1981). Fathering the chronically ill child. American Journal of Maternal-Child Nursing, 6, 124-128.

Meadow, K. P. (1968). Parental response to the medical ambiguities of congenital deafness. Journal of Health and Social Behavior, 9, 299-309.

Meadows, A. T., & Silber, J. (1985). Delayed consequences of therapy for childhood cancer. CA-A Cancer Journal for Clinicians, 5, 271-286.

Milburn, T. W., & Billings, R. S. (1976). Decision making perspectives from psychology: Dealing with risk and uncertainty. American Behavioral Scientist, 20, 111-126.

Mishel, M. H. (1981). The measurement of uncertainty in illness. Nursing Research, 30, 258-263.

Monat, A. (1976). Temporal uncertainty, anticipation time, and cognitive coping under threat. Journal of Human Stress, 2, 32-43.

Monat, A., Averill, J. R., & Lazarus, R. S. (1972). Anticipatory stress and coping reactions under various conditions of uncertainty. Journal of Personality and Social Psychology, 24, 237-253.

Montagna, P. (1980). Uncertainty as a scientific concept and its application to the study of occupations and organizations. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger Publishers.

Moos, R. H., & Billings, A. G. (1982). Conceptualizing and measuring coping resources and processes. In L. Goldberger & S. Breznitz (Eds.), Handbook of stress: Theoretical and clinical aspects. New York: Free Press.

Munsinger, H., & Kessen, W. (1964). Uncertainty, structure and preference. Psychological Monographs, 78, 586.

Norton, R. W. (1975). Measurement of ambiguity tolerance. Journal of Personality Assessment, 39, 607-619.

Okazaki, N. (1983). Life of advanced cancer patients after knowing the nature of their own disease: A personal experience of seven cases. Japanese Journal of Clinical Oncology, 13, 703-707.

Parkes, C. M. (1971). Psycho-social transitions: A field for study. Social Science and Medicine, 5, 101-115.

Parsons, T. (1980). Health, uncertainty and the action situation. In S. Fiddle (Ed.), Uncertainty: Behavioral and social dimensions. New York: Praeger Publishers.

Pearlin, L. I. (1983). Developmental perspectives on family and mental health. A report to the director of the National Institute of Mental Health by the Behavioral Science Research Review Consultant Panel.

Pearlin, L. I., & Aneshensel, C. S. (1986). Coping and social supports: Their functions and applications. In L. Aiken & D. Mechanic (Eds.), Application of social science to clinical medicine and health policy. New Brunswick, NJ: Rutgers University Press.

Peck, B. (1979). Effects of childhood cancer on long-term survivors and their families. British Medical Journal, 1, 1327-1329.

Perrin, J. M., & Ireys, H. T. (1984). The organization of services for chronically ill children and their families. Pediatric Clinics of North America, 31, 235-257.

Phillips, S., Bohannon, W. E., Gayton, W. F., & Friedman, S. B. (1985). Parent interview findings regarding the impact of cystic fibrosis on families. *Journal of Developmental and Behavioral Pediatrics*, *6*, 122-127.

Phipps, S. (1985-1986). The subsequent pregnancy after stillbirth: Anticipatory parenting in the face of uncertainty. *International Journal of Psychiatry in Medicine*, *15*, 243-264.

Pless, I. B., & Perrin, J. M. (1985). Issues common to a variety of illnesses. In N. Hobbs & J. M. Perrin (Eds.), *Issues in the care of children with chronic illness*. San Francisco: Jossey-Bass.

Poole, V. (1980). *Thursday's child*. Boston: Little, Brown and Co.

Rhoden, N. K. (1986). Treating Baby Doe: The ethics of uncertainty. *Hastings Center Report*, *16*, 34-42.

Roghmann, K. J., & Doherty, R. A. (1983). Reassurance through prenatal diagnosis and willingness to bear children after age 35. *American Journal of Public Health*, *73*, 760-762.

Rothstein, P. (1980). Psychological stress in families of children in a pediatric intensive care unit. *Pediatric Clinics of North America*, *27*, 613-620.

Sabbeth, B. (1984). Understanding the impact of chronic childhood illness on families. *Pediatric Clinics of North America*, *31*, 47-57.

Sabbeth, B. F., & Leventhal, J. M. (1984). Marital adjustment to chronic childhood illness: A critique of the literature. *Pediatrics*, *73*, 762-768.

Salk, L., Hilgartner, M., & Granich, B. (1972). The psycho-social impact of hemophilia on the patient and his family. *Social Science and Medicine*, *6*, 491-505.

Sandelowski, M. (1986). The problem of rigor in qualitative research. Advances in Nursing Science, 8, 27-37.

Sandelowski, M. (1987). The color gray: Ambiguity and infertility. Image, 19, 70-74.

Schuler, D., Bakos, M., Zsambor, C., Polcz, A., Koos, K., Kardos, G., & Revesz, T. (1985). Psychosocial problems in families of a child with cancer. Medical and Pediatric Oncology, 13, 173-179.

Schutz, A. (1970) On phenomenology and social relations: Selected writings (H. Wagner, Ed.). Chicago: The University of Chicago Press.

Shalit, B. (1977). Structural ambiguity and limits to coping. Journal of Human Stress, 3, 32-45.

Shapiro, J. (1983). Family reactions and coping strategies in response to the physically ill or handicapped child: A review. Social Science and Medicine, 17, 913-931.

Siegler, M. (1975) Pascal's wager and the hanging of the crepe. The New England Journal of Medicine, 293, 853-857.

Siemon, M. (1984). Siblings of the chronically ill or disabled child: Meeting their needs. Nursing Clinics of North America, 19, 295-307.

Stein, R. (1983). Growing up with a physical difference. Children's Health Care, 12, 53-61.

Stern, P. N. 91980). Grounded theory methodology: Its uses and processes. Image, 12, 20-33.

Strauss, A. L. (1975). Chronic illness and the quality of life. St. Louis: C. V. Mosby.

Strauss, A. L. (1987). Qualitative analysis for social scientists. New York: Cambridge University Press.

Suls, J. & Mullen, B. (1981). Life events, perceived control and illness: the role of uncertainty. Journal of Human Stress, 7, 30-34.

Terracini, B., Pastore, G., Zurlo, M. G., Masera, G., Fossati-Bellani, F., Castello, M. Tamaro, P., Massolo, F., Rosati, D., Biddau, P. F., & Russo, A. (1987). Late deaths and second primary malignancies among long-term survivors of childhood cancer: An Italian multicentre study. European Journal of Cancer Oncology, 23, 499-504.

Tversky, A., & Kahneman, D. (1973). Availability: A heuristic for judging frequency and probability. Cognitive Psychology, 5, 207-232.

Tversky, A., & Kahneman, D. (1974). Judgement under uncertainty: Heuristics and biases. Science, 185, 1123-1131.

Waddell, C. (1983). The process of neutralization and uncertainties of cystic fibrosis. Sociology of Health and Illness, 5, 210-220.

Waller, D. A., Todres, I. D., Cassem, N. H., & Anderton, A. (1979). Coping with poor prognosis in the pediatric intensive care unit. American Journal of Diseases of Children, 133, 1121-1125.

Wallsten, T. S. (1983). The theoretical status of judgemental heuristics. In R. W. Scholz (Ed.), Decision making under uncertainty. North Holland: Elsevier Science Publishers B. V.

Watson, J. (1985). Reflections on different methodologies for the future of nursing. In M. M. Leiniger (Ed.), Qualitative research methods in nursing. New York: Grune and Stratton, Inc.

Wiener, C. L. (1975). The burden of rheumatoid arthritis: tolerating the uncertainty. Social Science and Medicine, 9, 97-104.

Wolf, F. M., Gruppen, L. D., & Billi, J. E. (1985). Differential diagnosis and the competing-hypothesis heuristic. A practical approach to judgement under uncertainty and Bayesian probability. Journal of the American Medical Association, 253, 2858-2862.

Wright, G. N. & Phillips, L. D. (1980). Cultural variation in probabilistic thinking: Alternative ways of dealing with uncertainty. International Journal of Psychology, 15, 239-257.

Zimmer, A. (1983). Verbal vs numerical processing of subjective probabilities. In R. W. Scholz (Ed.), Decision making under uncertainty. North Holland: Elsevier Science Publishers B. V.

APPENDIX A

Topical Outline for Parent Interview

APPENDIX A

Topical Outline* for Parent Interview

Parents will be asked to discuss, as fully as possible, their experiences, understanding, beliefs, concerns, and management strategies concerning:

1. The period surrounding the diagnosis of the child's health problem.
    a. I would like you to tell me as much as you can remember about that period of time just before (child) was diagnosed.
    b. Who was it that first suspected that something might be wrong? What was the first cue? What did you think it meant? Did you have any uncertainty at <u>that</u> time about the meaning or the significance of the symptom(s)? Had there been other times before that (child/other children) had the same symptoms? How did you handle it before? Did you or someone else begin to get suspicious that this illness (symptom) was different in some way from previous similar illnesses? What did you do?
    c. When did you find out that (child) had (disease)?
    d. How long did it take to get a positive diagnosis? Was there any uncertainty (medical or in your own mind about the diagnosis?
    e. Do you remember what you were thinking and feeling while you were waiting for the diagnosis (results of tests)?
2. The etiology of the illness
    a. What were your thoughts or beliefs about what caused the illness?
    b. Have those thoughts or beliefs changed?
3. The initial, current, and future therapy and medical supervision
    a. How was your child's illness managed right after the diagnosis? Did you understand the purpose ofthe treatment(s)?
    b. What is currently being done to manage the illness?
    c. Do you know what kind of therapy (if any) will be needed in the future?
4. The predictability (day-to-day and long-term) of their child's health status
    a. Does your child's health problem ever make it difficult to plan for social or family events?
    b. Make financial plans for the future?
5. The parents' ability to judge and make decisions regarding the child's behaviors (normal/developmental vs illness)
    a. Can you generally tell if your child is doing okay or do you find that you are unsure much of the time?

- b. Do you feel comfortable deciding when your child just has a minor illness, such as a cold, and when he/she needs to see a doctor?
- c. How comfortable do you feel managing (child's) ordinary or common illnesses compared to your other children's?
- d. Does (child's) condition vary from day to day (week to week, month to month)? Do you feel comfortable in managing these fluctuations on your own?
- e. Do you ever worry that you will miss an important symptom and not take child to doctor when you should? Has this ever happened?

6. Awareness of alterations in parenting behaviors since the diagnosis
   - a. Has (child's) illness changed the way you treat him/her? Or the way you treat (child's) brother(s)/sister(s)?
   - b. Has it changed the way you behave as a parent or feel about your parenting skills?
7. Alterations in behaviors of other family members or the family as a unit since the diagnosis
   - a. How do you think (child's illness has affected your family as a whole?
   - b. Can you identify specific areas of stress that it has created? How do you manage this stress (what strategies do you use)?
8. Future-oriented plans
   - a. When you think of the future, what kinds of concerns do you have for (child)/family?
9. Phase related stressful events
   - a. Could you describe for me what you remember as being the most difficult for you during each of these periods?
     - Pre-Illness/Pre-Diagnostic
     - Diagnostic
     - Early Treatment
     - Continued treatment
     - Discontinuation of treatment 10. Uncertainties
   - a. What kinds of uncertainties have you experienced related to (child's) illness.
   - b. Describe the strategies you use to manage.

* The investigator will conduct the interview in an unstructured manner, introducing the topics and encouraging the parents to proceed with their narrative in a way that best represents their experiences. Topics of particular interest to the study are outlined above, along with some representative questions that might be asked. Probes will be used to help the parents clarify or expand a statement where indicated.

APPENDIX B

Recruitment Letter to Parents

APPENDIX B

Recruitment Letter to Parents

Dear (Parent):

I am writing to ask you to take part in a study of families who have a child with some type of chronic health problem. This study is trying to learn more about the kinds of situations that create or reduce the feelings of uncertainty that parents often have as a result of their child's chronic illness. This information will be used to help health professionals provide useful counseling to parents and others who work with children with a chronic illness.

As part of this study, I would like to interview you for about one to two hours in your home. I will be asking about such topics as how you manage your child's illness at home, how you make decisions regarding the need to have your child seen by a doctor, and the ways that your child's illness has affected your family's planning of day to day activities.

The families being interviewed for this study were referred by doctors, hospitals and clinics. Your name was referred to me by (insert name). Within the next few days I will call to ask for an appointment at a time that will be convenient for you. If you have any questions, I will be happy to answer them when I call, or you may telephone me at 476-9494. If you do not want me to contact you to inquire about your interest in participating in the study, please fill out the enclosed postcard and return it to me.

The information that you can provide will be useful in developing ways for health professionals to better assist families whose child has a chronic illness. Findings will be in a form in which no person or family can be identified, and the confidentiality of your responses will be protected.

Thank you for your cooperation.

Sincerely,

Marsha H. Cohen, R.N., M.S.
Doctoral Candidate
Department of Family Health Care Nursing

APPENDIX C

Consent To Act as a Research Subject

APPENDIX C

CONSENT TO ACT AS A RESEARCH SUBJECT
in
A Study of Coping Responses of Families
to Sustained Uncertainty

Marsha Cohen, R.N., a doctoral candidate at the School of Nursing, is doing a study to learn more about the ways that families manage the many predictable and unpredictable situations that occur as a result of having a child with a chronic health problem. Because I have a child with a chronic illness, I have been asked to participate in this study.

If I agree to participate, I will be interviewed in my home for one to two hours. With my permission, the interview will be audiotaped so that my responses can be accurately and completely recorded. I will be asked about the situations that create or reduce my feelings of uncertainty concerning my child's health, how I make decisions regarding my child's need for medical attention, and how my child's illness has affected my family's day-to-day activities. In addition, if I consent, the investigator may contact me at a later date to clarify or verify the findings of the study in order to insure that it represents an accurate accounting of my experience.

As a result of answering these questions, there is a possible loss of my privacy. The investigator will separate names from responses and will keep the names coded and the code locked. The audiotapes will be erased after the analysis of the data has been completed. Individual data will not be released to anyone or to any participant. The confidentiality of my responses will be protected as fully as possible under the law.

There will be no direct benefit to me or my family from participating in this study. The investigator hopes to learn more about the needs of families of chronically ill children in order to help health professionals provide useful counseling and other assistance to parents who have a child with a chronic illness.

My participation is voluntary. I may refuse to participate or I may decide at any time that I do not want to continue, and the interview will be stopped. I may also chose not to answer a particular question but to continue with the interview. I may withdraw from the study at any time and my decision to withdraw will not affect or influence my child's continued medical care.

I have talked with Ms. Cohen about this study and all my questions have been answered. If I have other questions, I may call Ms. Cohen at 476-9494. I have received a copy of this form and the Experimental Subjects' Bill of Rights to keep.

______________________________	___________________________
Investigator's Signature	Respondent's Signature
	___________________________
	Date