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Outcomes of Pregnancy in Women with Cystic Fibrosis

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A dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy in Nursing at Virginia Commonwealth University.

by

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Dedication

Remember This My Child

I tried to do the best I could with what I had
while I watched you
seemingly unburdened
moving effortlessly through time.

I will always see you
growing
playing
dating
college
marriage
children
freely savoring the many joys of life.

Unaware that I
can never experience these joys
in the same way
with each passing year
my joy, my hope
intermingles with despair
more symptoms
more exacerbations
more treatments
always tired
But desiring always to be present in your life.

The cost to me of all these things
of even you my child
could be my own life
my hope remains always
to see you
experience you
savoring the many joys of life
watching you as you grow
ever present.

But always know
always remember
I tried to do the best I could with what I had
and the best was you.

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ABSTRACT

OUTCOMES OF PREGNANCY IN WOMEN WITH CYSTIC FIBROSIS

By Bethany Jean Geldmaker, R.N., P.N.P., M.S.N.

A dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of Philosophy at Virginia Commonwealth University, 2000.

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Medical advances in disease management has improved quality of life for women with cystic fibrosis (CF) and now enables them to consider pregnancy. The purpose of the study was to examine women's perceptions of how the demands of having CF influenced their ability to care for their children and continue to meet their own health needs. The study design was grounded theory methodology with a complementarity research technique to incorporate qualitative and quantitative data collection. Participants were 12 women recruited over the Internet and through an adult CF newsletter who completed questionnaires for demands of illness (DOII) and self care of CF. A severity of illness measure (NIH-CSCF) based on medical records was completed by participant's health care providers. Participant interviews were conducted following receipt and preliminary analysis of study instruments. Semi-structured telephone interviews incorporated information from questionnaires. A data matrix containing information from questionnaires and the participant interview was compiled for each participant. Within and across data matrix analysis elucidated patterns which were further condensed into categories. A theoretical model emerged that reflected social, medical, and economic themes of how women worked to sustain and maintain family life and a chronic illness entering the twenty first century. The findings also provided recommendations for health care providers working with mothers with CF.

Chapter 1

Introduction

The practice of medicine in the treatment of chronic illness in past decades was limited in what could be done for patients. Chronic illness, as with other aspects of culture, reflected social mores situated in time. Historically, a montage sprang up around the culture of illness. This was reflected in literary works and paintings that depicted pale, lifeless figures reclining in death-like poses. Doctors primarily were engaged to make patients comfortable and provide support to families.

The aim of health care in the twenty-first century for individuals with chronic illness is to enhance and promote health despite inevitable, and often unpredictable, fluctuations in functional status. One of the great triumphs of American health care is that technological advancements are changing the nature, and subsequently the culture, of chronic disease. For example, survivorship for children with cystic fibrosis (CF) two decades ago was adolescence (Davis, Drumm, & Konstan, 1996). Children diagnosed with CF at the end of the twentieth century have an average life expectancy of up to 31 years of age (Cystic Fibrosis Foundation, 1997).

The lag between medical advances and societal norms and expectations has been reflected in adults with CF. This lag has posed two major challenges for adults with CF. Traditionally, little emphasis has been placed on preparing adolescents with CF and their families for transition to adulthood. The second challenge has been with pregnancy and child rearing as a mother with CF.

Increasing length of survivorship means that individuals with CF now experience maturation into adulthood. Recent progress in medical technology has enabled individuals with what was once a fatal childhood disease, to experience challenges and responsibilities of young adults. Such changes in quality of life for individuals with CF, have increased opportunities for independent living, formation of intimate relationships, and consideration of pregnancy. This process has been a challenge for young adults with CF, one for which they generally have not been prepared.

In particular, women with CF are making decisions about pregnancy. Ironically, this period of time which traditionally is the healthiest and most productive for young adults, marks potentially the terminal phase of a woman's life with CF. Decisions about pregnancy are done with the knowledge that they may not live to see their children grow-up nor survive the pregnancy. The outcome, both in terms of health and adjusting to pregnancy and parenthood, is poorly understood.

In summary, change in life expectancy for women with CF brings renewed hope. It means that for the first time opportunities for a life independent of parents, inclusive of career, marriage, and significantly, children, are possible. Normal developmental tasks must be dealt with in addition to potential changes in functional health status and the realization that death is not imminent. Little is known about outcomes of child rearing and disease management in women with CF.

Purpose

The purpose of this research was to examine what demands chronic illness places on women with CF as they attempt to care for themselves and children. The specific aim is to understand how women with CF incorporate their disease and its treatment during pregnancy and with the change in responsibilities following the birth of a child.

Research Questions

The following questions were addressed:

1. How was CF affected during pregnancy?

2. How was CF affected following the birth of a child?

3. Are there demands placed on women with CF during pregnancy that affect their ability to care for themselves?

4. Are there demands placed on women with CF following the birth of a child that affect their ability to care for themselves?

5.What supports are needed to help women with CF during and after pregnancy deal with illness responsibilities?

6. What supports are needed to help women with CF care for their children?

Definitions

The following conceptual definitions served as a guide for the research:

1. Cystic fibrosis is an autosomal recessive disease caused by mutations of a gene located on the long arm of chromosome 7 (Stern, 1997). The gene product in the cystic fibrosis transmembrane conductance regulator (CFTR) is a protein that regulates and participates

in the transport of electrolytes across epithelial-cell membranes. The manifestations of the disease are presumed to be related to abnormal transport of electrolytes and include respiratory, gastrointestinal, skin, and reproductive tracts (Davis et al., 1996; Ramsey, 1996). Diagnostic testing is done primarily through the determination of sweat chloride concentrations, and by genotyping, usually initiated by characteristic clinical findings (specific gastrointestinal and pulmonary disease) and less typical clinical characteristics (nasal polyps, pancreatitis, congenital absence of the vas deferens/obstructive azoospermia, and pansinusitis) (Stern, 1997).

2. Chronic illness is a multidimensional experience that places a variety of demands on individuals. The perceptions of the demands of related illness events are generated by the disease and its treatments.

3. Demands of illness are the challenges individuals face in adjusting to life with a variety of related illness demands (Packard, Haberman, Woods, & Yates, 1991). These challenges may include the direct physiologic effects of disease and personal disruption that occurs as a consequence of the disease.

Philosophic Traditions

Symbolic interactionism provided the philosophic foundation for this study. The underlying fundamental element of symbolic interactionism is that man must be studied on his own level. The predilection is to stay close to the world of everyday experience from which man's viewpoint developed and with which it sought to deal.

The philosophic roots of symbolic interactionism extended back to the eighteenth century and to Scottish moral philosophers, such as David Hume, Francis Hutcheson, Adam Smith, and Adam Ferguson. For these scholars, the object of inquiry was understanding events of ordinary man as a way to understand future regulation of his conduct (Bryson, 1945). As a result of their writings, the treating of man as a natural object and the scientific study of everyday experience were legitimized.

Symbolic interactionism is a world view that provides a framework for how people make sense of their life-situations and the ways in which they go about their daily activities. It is grounded in the notion that the study of human lived experience must be accomplished through the day-to-day practices and experiences of human group life of the population of interest. Symbolic interactionism was primarily developed by George Herbert Mead and Herbert George Blumer (Blumer, 1969; Stryker, 1980).

Symbolic interactionism asserts several important axioms. The first of these is that social behavior is the product of a role-making process and that the enactment or performance of a role is variable. It asserts the notion of choice in performing a role. With choice comes the opportunity to reject the expectations attached to a position occupied or to modify the performance called for in a situation. The person is seen as a structure of positions and roles, which internalized, is the self. The mutually determinative relation of self and society is basic to symbolic interactionism.

The second axiom is that interactions of person and society are patterned and organized and are derived primarily from interaction. These interactions are a mosaic of

interdependent and highly differentiated parts. As a result, not all behavior is describable in terms of structure of positions and roles.

Third, human beings act toward things on the basis of meanings those things have for them. Meaning is the product of interaction. Subsequently, meanings are processed and modified through an interpretative process with each encounter.

Lastly, humans create and use symbols (Blumer, 1969). Communication occurs as a result of symbols. Role-taking involves interaction through the act of interpretation of symbols emitted by others. The act of interpretation allows the emergence of the capacity to interact between humans and forms the basis of society (Prus, 1996). Response is based on meaning and interpretation attached to actions, not on the actions themselves. Role-taking enables man to indicate oneself to things in the world, create objects, provide meaning to objects, and to guide action by what was noted. Action is the product of piecing together and guiding one's own action, by taking account of things and interpreting their significance for prospective action.

In summary, the human infant enters life as neither a social nor an antisocial creature with the potential for social development (Stryker, 1980). Humanness develops as a result of interaction with and interpretation of social processes. Consequently, it is possible to study man's social behavior through an analysis of the culture or society in which he resides.

Methodologic Framework

Grounded theory, as interpreted by Glaser (1978), was used as the methodologic framework through which the social reality of participants was constructed. Certain fundamental principles emerged from philosophic and theoretical traditions that were key to grounded theory. Grounded theory method is rooted in symbolic interactionism in an attempt to derive meaning from symbolism attached to the participant's world (Stern, 1994).

Inherent in the practice of grounded theory method is the researcher-as-human-instrument with immersion in the participant's world. The researcher entered into active dialogue with participants and worked to maintain a hermeneutic dialogue and circle with participants. Analysis of human action was located or situated within a social context.

Grounded theory method connects the multiplicity of interpretation and perspective of participants with that of the researcher (Strauss & Corbin, 1994). This approach employs certain assumptions about the world, the people in it, and the proposed research. These assumptions included the following: that meaning was expressed verbally, in action, and practice; that to understand human behavior one needed to look at everyday practices; that participants were meaning-giving beings; that meanings were contextually based; that meaning and significance of human action was fluid and changing; that interpretation was necessary to understand action; and that facts were value-laden (Addison, 1992).

The intent of research questions within grounded theory method was to gain insight into 1) the perception of the demands of the disease in women with CF during pregnancy and following the birth of a child, and 2) how the disease influenced the transition to becoming a parent. This intent precluded sole use of a single method and research design. A synthesis of styles was used as part of the multi-method approach found with grounded theory framework.

Limitations, Delimitations, and Assumptions

The strength of grounded theory method was the multi-method approach for discovery and voice given to participants. However, the emphasis of focus and context represented only the experiences of women included in the research study. The findings were not intended to be generalized to other mothers with chronic illness.

The issues of meaning and interpretation by participants in construction of their social reality were central to grounded theory method. Descriptive and interpretive validity of data reported by respondents was assessed on the validity of how accounts related to those things which women's accounts claimed to be about (Maxwell, 1992). In other words, the applicability of the concept of validity did not depend on absolute truth or reality to which an account could be compared. Rather, it was to be assessed relative to purposes and circumstances—that understanding was the fundamental concept for construction of interactants' social reality.

A multi-method approach was more than the combination of different kinds of data to produce a more complete picture (Brewer & Hunter, 1989; Fielding & Fielding, 1986).

Combining different methods added range and depth, but not necessarily accuracy. Use of multiple sources of data without bias-checking procedures may have served to multiply error.

In summary, use of multiple strategies for data generation and analysis was supported by grounded theory method and congruent with the philosophic purpose and goals of the method (Leininger, 1994). The researcher was investigating what symbolic meaning emerged for women with CF as they grappled with having a chronic illness, being pregnant, giving birth and learning how to care for a child. Through integration of physiologic data and perception of participant's ability to deal with the demands of illness, pregnancy, and children, the investigator was attempting to construct what interactants saw as their social reality.

Conceptual Framework

Transition theory, as described by Parkes (1971), was the conceptual framework used to guide the study. Transition theory accommodates continuity as well as discontinuity associated with multiple transitions. For women with CF this included potential changes in severity of illness during and following pregnancy, living with chronic illness, and learning to care for a child.

Definitions

A life transition begins when there is disruption in reality or when disruption necessitates restructuring of the existing reality (Selder, 1989). Discontinuity occurs with a life transition that is associated with a shift in previous life events and the emergence of

coping skills in an attempt to re-establish stability and continuity (Chick & Meleis, 1986; Hopson & Adams, 1977). Life space consists of those parts of the environment with which the self interacts and in relation to which behavior is organized (Parkes, 1971). It is the mind and body with which the self interacts.

Transition Theory

Transition requires major reorganization of life space regardless of whether the focus is on individual, group, or organization. The importance of changes in life space are influenced by assumptions individuals make about the world. The total set of assumptions created within individuals compose what Parkes called man's "assumptive world" (Parkes, 1971). The assumptive world is based on past experiences and forms from a constellation of sensations. The constellation is part of the assumptive world and enables individuals to recognize part or whole of situations in the present and to make correct predictions about the characteristics and utility of present experiences. The assumptive world includes everything known or thought to be known. It includes interpretation of the past and expectations for the future. Life space constantly changes as fresh combinations of communications are received and assimilated.

Whenever major life changes occur, individuals restructure their life space. Major changes in life space reveal gaps in expectations of the future based on past models of response (Parkes, 1971). The ability to recognize familiar objects and to orient oneself within an environment involves mastery of the environment regardless of the extent to which the situation is actively controlled or altered.

Transition theory helps to link the interplay between person and environment. It serves as a framework by which to represent the constellation of patterns and responses over time that are embedded in context and situation (Chick & Meleis, 1986; Schumacher & Meleis, 1994). Transition conditions incorporate several influencing factors: meanings, expectations, levels of knowledge and skill, level of planning, and emotional and physical well-being (Schumacher & Meleis, 1994).

The experience of transition consists of three phases: entry, passage, and exit (Brammer, 1991; Bridges, 1991). The transition period is characterized by process, disconnectedness, perception, and patterns of response. The disruption and disassociation that are associated with pre-transitional state are countered on successful completion of transition (Bridges, 1991; Chick & Meleis, 1986; Selder, 1989). The completion of transition implies that a person has reached a period of less disruption or greater stability.

Subsumed under the broad umbrella of transition theory, is the transition that occurs through daily living with a chronic disease process (Shaul, 1997). The nature and intensity of demands that women with chronic illness encounter are thought to dictate this response and adaptation to these demands. This research sought to explore the validity of this assumption in women with CF.

For women with CF, transition is not always under voluntary control. Women with CF experience transition in both developmental and biological health. Adolescent girls with CF experience more physical maturational delay and more rapid decline in pulmonary status than boys. There seems to be an association of pulmonary decline with advanced

Tanner staging, age of acquisition of Pseudomonas aeruginosa, and in adequate nutrition (Cromer et al., 1990; Cystic Fibrosis Foundation, 1997; Johannesson, Gottlieb, & Hjelte, 1997).

The association of health and illness to a life transition in chronic illness is not well understood. Cowan (1991) theorized that biological functions may be involved in determining timing, duration, and adaptational outcomes of transitional life events. Whether patients experience more disease exacerbations around major life transitions is unknown. Outcomes of transition are thought to perpetuate uncertainty for individuals with a progressive chronic illness (Brown & Powell-Cope, 1991; Corbin & Strauss, 1988; Loveys, 1990).

Demands of illness.

Under the umbrella of transition theory is included a construct called the demands of illness. The construct, demands of illness, was developed by Woods, Haberman, & Packard (1993) to examine responses of families and individuals to chronic illness. Within this construct, 3 domains are hypothesized to emerge in response to a chronic illness. These include: (a) disease-related demands, (b) personal disruption demands, and (c) environmental transactions. The types of illness demands and coping responses that are evoked represent different phases of chronic illness (e.g., early post-diagnostic phase). The ability to characterize a developmental course of family response to the experience of chronic illness fits within a transitional framework. The notion of a differential relationship

of illness demands and individual, dyadic, and family adaptation supports research of transition in mothers with CF.

Medical advances have enabled survival of women with CF into adulthood. Interest in marriage and/or a family has been increasing for women with CF; however, women's health may be less than optimal for childbearing. Adolescence may produce a decline in pulmonary function for women with CF. Questions about pregnancy and care for children of women with CF based on an uncertain life expectancy, raises important ethical and moral questions. How should decisions be made about pregnancy and children--through psychological egoism, which primarily considers the individual's desire to bear children despite potential negative outcomes, or through the principle of utility in which the best consequences for everyone concerned takes precedence (Rachels, 1993)? Ethical and moral concerns also exist about outcomes of quality of life and family functioning after pregnancy.

Passage from one life phase to another incorporates elements of process, time, and perception. The purpose of transition is structure to create new meaning where old meaning has been disrupted. The outcome of transition may be affected by physical and social demands, availability of resources, and meanings attached to transition. For women with CF, little was known about the experience of pregnancy and parenting and the role the demands of their disease had during and following pregnancy.

Summary

Existing theory about the outcome of child bearing and rearing was incomplete for women with CF. Grounded theory method permitted the use of several approaches to data generation and analysis. These multiple approaches to data generation allowed more complete exploration of the phenomenon of interest. A more complete exploration of the phenomenon in turn serves to improve existing models or to develop alternative models to fill gaps in transition theory.

Chapter 2

Existing Theory

The philosophic roots for this study were grounded in symbolic interactionism. Transition theory was the overall conceptual theory because it supported the notion of the study of human lived experience by examining the social interaction, meaning, and interpretation an experience has for individuals. These foundations permitted the development of a framework based on social interaction of how people strive to make sense of their world.

Historically, children faced with the diagnosis of CF were not expected to survive beyond adolescence. The illness culture consisted of daily complicated therapies primarily aimed at comfort. However, dramatic changes in medical therapy in the past decade has enabled survival beyond adolescence, thereby shifting socio-cultural expectations.

The need for greater understanding of reproductive health needs in women with CF has been emphasized by an increasing frequency of single reports of pregnancy. The first case report appeared in 1960 (Siegal & Siegal, 1960). To date, little data exists as to prevalence or outcomes of pregnancy in the CF population. Individuals with severe lung disease were most likely to develop complications, however, studies consisted of case reports of very small samples (Edenborough, Stableforth, Webb, Mackenzie, & Smith, 1995; Frangolias, Nakielna, & Wilcox, 1997; Hilman, Aitken, & Constantinescu,1996; Kent & Farquharson, 1993).

Pre-pregnancy lung functions were found to be a useful predictor of outcome measures in pregnancies in women with CF. However, guidelines used to advise women about pregnancy are based on case reports and surveys and use of retrospective data. Despite the accepted view that pregnancy is less hazardous in women with milder disease, a paucity of data exists on long-term outcomes of pregnant versus nonpregnant women with CF (Edenborough et al., 1995; Frangolias et al., 1997; Hilman et al., 1996; Kent & Farquharson, 1993).

The decision to have children, in terms of outcome for women with CF and their families, has far reaching implications. Chronic illness may make it difficult for women to perform certain parenting and family tasks depending on severity of illness. Because the burden of pregnancy and child rearing tends to fall disproportionately on women, the focus in this research was on females.

The challenge of being both patient and parent, of raising children when one has a chronic illness, has not been extensively examined in health care literature. Two areas were explored related to being a mother with a chronic illness: (a) maintaining functional health status--caring for themselves and their illness, and (b) caring for children.

Maintaining Functional Health Status

Understanding issues regarding functional health status is important for the development of interventions to maintain or improve quality of life. Functional health status in women with CF and other chronic illnesses, includes: (a) outcomes of pregnancy, (b) coping with the demands of illness, and (c) life expectancy and end of life decisions.

Outcomes of pregnancy.

The decision to become pregnant, in terms of outcome for women with CF and their families, has far reaching implications both ethically and morally. Concerns about quality of life, life expectancy, and fetal exposure to CF medications during pregnancy have been raised (Hilman et al., 1996; Kent & Farquharson, 1993). Outcomes and problems of pregnancy may persist beyond delivery. Maternal mortality figures were reported to be 18% within 24 months of delivery and of those, 12% died within 6 months of delivery (Cohen, di Sant'Agnese, & Friendlander, 1980; Corey, Levison, & Crozier, 1976; Palmer, Dillon-Baker, Tecklin, 1983). Recent epidemiologic figures report average CF life expectancy to be 31.3 years and the age range in CF pregnancy to be 15 to 39 years (M=22.6 years) (FitzSimmons, Willie, & Fiel, 1995; Hilman et al., 1996; Cystic Fibrosis Foundation, 1997). Based upon these statistics, women may experience less than a decade of life following pregnancy.

Demands of illness.

Despite the challenges associated with self-care and CF, 1996 statistics for 6,424 adults 18 years and older in the Cystic Fibrosis Foundation Registry (1997) found that 36% were employed full-time, 13% part-time, 22% unemployed, and 24% were students. Of this group, 62% were single and 29% were married. Sixty-one percent had completed high school and 30% completed college or graduate school. While secondary databases provide large sample sizes, they are limited in the detail and type of information that can be obtained.

Studies examining adjustment of living with a chronic disease have primarily focused on psychological adjustment to disease and have not examined other issues of transition to independent living. Additionally, studies have dealt with acquired illnesses such as cancer, HIV/AIDS, and arthritis (Hackl, Somlai, Kelly, & Kalichman, 1997; Packard et al., 1991; Sidell, 1997). For children with CF, families have had to learn to deal with cyclical crises and issues of long term survival (Cowen et al., 1984; Lampo, Dab, & Malfroot, 1990). How families and adult patients achieve balance between the demands and limitations of CF is only beginning to be explored.

Life expectancy.

Recently, survival in CF has been found to be linked to gender. Mortality figures were examined for both sexes between 1988 to 1992 and revealed survival in females aged 1 to 20 years to be poorer than males (relative risk = 1.6; n = 21,047) (Rosenfeld, Davis, FitzSimmons, Pepe, & Ramsey, 1997). While the average life expectancy was 31 years in general, the median survival in years was 25.3 for females and 28.4 for males. Outside this age range, male and female survival rates were not significant.ly different. The only factor that seemed to differ between sexes was earlier colonization by females of Pseudomonas aeruginosa. More studies on airway inflammation and infection are needed to further explore difference in gender effects at different ages.

In summary, few studies have examined outcomes of pregnancy and demands of illness from the perspective of women with CF. A paucity of data about quality of life and maintaining functional health status exists to date.

Issues of Parenting--Caring for Children

Life transition following pregnancy, involves role changes in position, shifts in family and peer loyalties, and a change in entitlements and obligations with family and society (Baumrind, 1991; Golan, 1981; McAnarney, 1985). Changes that occur stimulate disequilibrium. For women with a chronic illness such as CF, there are many factors that influence their ability to care for their children. Discussion will center around the following: (a) family functioning--disease stage and family adaptation, and (b) social support.

Family functioning.

Achieving independence from family has not been easy for individuals with CF. Chest physiotherapy (CPT) for removal of lung secretions occupies several hours per day. Boyle, di Sant' Agnese, Sack, Millican, and Kulcyzcki (1976) found that while spouses of women with CF had been informed prior to marriage of the nature of the disease, they were unprepared for complex treatment regimens and repeated crises.

Independent functioning for individuals with chronic illness depends on the ability to perform tasks related to caring for self, caring for children (parenting), and controlling disease symptoms. Physical functional status in 20 mothers with narcolepsy was examined by Nehring and Cohen (1995). Response items were scored on a 40 item scale of difficulty from 0 to 3 on perceived severity of illness, degree of quality of life effects, and parenting tasks. Symptom management pervaded the family's adjustment to chronic illness. Events

viewed as problematic were those that occurred during the mother's most symptomatic period of the day.

Concerns about family functioning of women with breast cancer was examined by Gotay (1984). Concerns were found to be different based on phase of illness, disease transition, and perception of family functioning. These findings were in part supported by Woods et al. (1993) who found that women's perceptions of demands of the health care system and family adaptation were related to post-diagnosis, illness exacerbation, or terminal phase of their breast cancer. The intermediate phase of illness reflected different concerns based on controlling symptoms which persisted throughout the course of the illness.

Similar issues were raised in a study of single and married women with illnesses characterized by frequent exacerbations (e.g., rheumatoid arthritis, progressive systemic sclerosis, inflammatory disease). Thorne (1990) conducted extensive interviews with 16 mothers with chronic illness. Four thematic categories emerged: performance, availability, dependence, and socialization issues. Physical problems with fatigue and mobility, unpredictability of health, dependence on children for provision of care, and reliance on children for moral support were major sources of concern. Mothers reported frequently postponing their own health care when possible. The desire to maintain a normal home environment with few interruptions in routine was dominant in the interviews.

The postponement of care as a means to preserve family functioning, was found in another study. Custodial care for children was found to be the primary concern in

interviews with eight HIV-positive women (Hackl et al., 1997). Fear of social isolation because of the nature of their disease, as well as lack of resources socially and financially, created many difficult barriers. Secrecy regarding HIV diagnosis required hiding or refusing medication to avoid questions about health. The importance of the use of silence regarding diagnosis was perceived as a way to maintain an air of normality, to maintain a sense of parental status and dignity, and to best protect their children's well-being.

Other studies refuted the use of silence or isolation as an effective means of family functioning (Bloom, 1982; Lewis, Woods, Hough, & Bensley, 1989; Primomo, Yates, & Woods, 1990; Woods & Lewis, 1995). Familial introspection was found to be protective of family functioning. Introspective behaviors involve family reflection on their functioning and negotiation of future directions (Primomo et al., 1990). Measures of high levels of family cohesion and positive adjustment suggested that introspection within the family enhanced family functioning (Bloom, 1982; Primomo et al., 1990; Woods & Lewis, 1995). Long term consequences of chronic illness shifted family functioning; over time, social resources outside the family became less influential and internal relationships more important (Primomo et al.,1990).

Social support.

Social support, internal or external to the family, involves key people identified by the mother to assist her through illness and family demands. Various dimensions of support in women's support networks were found to be related to different types of adjustment to chronic illness (Hough, Lewis, & Woods, 1991; Shaul, 1997; Woods,

Yates, & Primomo, 1989). Women perceived more support from their partners than from any other source. The perception of affect, affirmation, and reciprocity from partner and family, resulted in less reported depression, higher marital quality, and better family functioning.

An important concern about reliance on family members for care and support was with the use of care givers under the age of eighteen years. The majority of patients with CF are young, dependent on parents for support and care, and living within an intergenerational family. No empirical studies exist about youngsters providing care to adults with CF. A limited number of studies have begun to raise awareness of children providing caregiving to adults with chronic illness (Aldridge & Becker, 1993; Gates & Lackey, 1998). Relevant issues that are raised with non-adult caregivers are the extent of responsibility requested and interference with normal developmental needs.

In addition to social supports based on human relationships, families frequently need outside services to maintain the mother's health. During disease exacerbation, problems arise in accommodating the mother's needs as well as caring for children. Professional and community service delivery was reported by mothers to be fragmented and unable to provide both clinical treatment and family support (Hackl et al., 1997; Nicholson, Geller, & Fisher, 1996; Peters & Esses, 1985; Thorne, 1990). Only when families were in crisis were they assessed as demonstrating need. Because of lack of adequate data, costs to individuals, families, and society are unknown.

In summary, findings from studies suggest that mothers with chronic illness face a complex set of challenges. Management of disease is at times at odds with the needs of the family. Resources necessary for family functioning and adaptation seem to cycle depending on severity of illness. Research examining family functioning has focused heavily on acquired diseases and intact families. Additionally, sample size and differences of measurement in research instruments make it difficult to generalize across studies. What has emerged is the importance of social support in family adaptation of mothers with chronic illness.

Summary

There is a growing body of knowledge about women with chronic illness. As technology improves the prognosis for many once fatal diseases, more women are opting to become pregnant. Research has primarily focused on illnesses such as HIV, arthritis, and cancer. While some comparisons may be made, the contrasts are far more profound. Women with CF have dealt with the consequences of their disease from the time of diagnosis, usually in early childhood. While CF begins as an invisible disease, it transitions into a visible one as evidenced by significant work of breathing, supplemental oxygen requirement, and activity limitations. Even with all the limitations imposed by their disease, women with CF attempt to establish new social and family relationships apart from their immediate families.

Comparisons with other studies on chronic illness in women can be made. Women frequently reported exchanging professional career trajectories for what might be

described as an illness career. A number of theories have been used to examine chronic illness, pregnancy, and caring for children. Default nursing concepts of family functioning, social support, family adaptation, and so forth, may be uniformly applied but do not serve to further illuminate new information of the experience. What has been lacking in other studies has been the development of new ways to examine chronic illness. Past studies have focused heavily on the perception of the disease, then identified patterns or themes, but prematurely stopped analysis without fully integrating or developing theory.

In summary, outcomes of pregnancy, the desire to be independent, and the impact a mother's chronic illness has on her ability to care for her children may lead to role conflict--that of patient versus parent. Whenever a major shift or change occurs, the need arises for restructuring ways of looking at the world and living in it. Old patterns of thought and activity must be reordered to accommodate the change. How a woman accommodates demands of disease in caring for herself and her family is only beginning to be examined in CF.

Chapter 3

Original Research Proposal Design and Methodology

It has been said that the best laid plans of man or woman sometimes "go astray" and such was the case with the initial phase of data collection for this study. Several unanticipated problems arose despite careful pre-proposal assessment and planning. The problems that arose were significant enough to require a reconsideration of the proposal and ultimately, recommendations for revision.

Revisions in the proposal also necessitated changes in the format of chapters in the dissertation. What follows in chapter three are the originally proposed methods that were approved initially by the dissertation committee. Chapter three concludes with a discussion of the emergent issues that arose during data collection. Chapter four describes the revised research methods that were actually used to collect and analyze data and closes with research questions that were answered using the revised methods. Chapter five presents the results of data analysis and chapter six concludes with a discussion of findings and their implications.

Design

The original design of the research proposal was grounded theory method, as interpreted by Glaser (1978). The intent of research questions posed in this study was to gain insight into the perception of demands of the disease placed on women with CF during pregnancy and following the birth of a child and the influence of the disease on the transition to parenthood.

The study design was cross-sectional and reflected that point in time during which questionnaires and interviews were completed by participants. Of interest were participants' perceptions of health behaviors contextually based on events leading up to the present. The study was not designed to predict future health and health-related behaviors or to measure health outcomes. The expected outcome was a theoretical model that would describe and explain the relationships among disease, pregnancy, and child care demands in women with CF. Research questions were aimed at exploring the relationship between CF, pregnancy and care of the child, and care of the disease. The proposed research questions were:

1. How was CF affected during pregnancy?
2. How was CF affected following the birth of a child?
3. Are there demands placed on women with CF during pregnancy that affect their ability to care for themselves?
4. Are there demands placed on women with CF following the birth of a child that affect their ability to care for themselves?
5. What supports are needed to help women with CF during and after pregnancy deal with illness responsibilities?
6. What supports are needed to help women with CF care for their children?

The use of grounded theory method was chosen to allow for probing of tacit knowledge of respondents and to document changes in physiologic measures of CF (Dzurec, & Abraham, 1993; Morse, 1991). The goal in using a combination of data

sources and analysis was not convergence but rather to examine a variety of influences on the phenomenon of interest. By fostering interdependence and interaction of data, comparisons as well as sharp contrasts of research findings were expected.

Sample

The population of interest were women with CF who were pregnant or who had been pregnant and had given birth to one or more living children. The sample was to be drawn from CF Centers in the United States. Selection of centers was to be based on: (a) their willingness to provide access to women who were pregnant or who had been pregnant and had children; and (b) had accreditation through the CF Foundation.

Sampling was to begin with women who were pregnant or who had been pregnant and had a living child. All women who agreed to participate in the study were to complete a Demands of Illness Inventory (DOII) and Self-Management Behavior Form (SMQ-CF). A severity of illness measure, the National Institutes of Health-Clinical Score for CF (NIH-CSCF), was to be obtained on each women by the investigator. The projected sample size was 140 based on pre-proposal reports of the availability of participants (Knapp & Brown, 1995).

The next level of sampling was to be selective and theoretical for participant interviews (Glaser, 1978). Selective sampling of women was to be based on analysis of their responses to the DOII, SMQ-CF, and NIH-CSCF. As the theory developed during on-going constant comparative analysis, theoretical sampling was to continue in order to confirm or disconfirm conditions under which the developing theoretical model was

applicable (Miles & Huberman, 1994). The number of interviews was to be based on maximum variation of participant responses in order to adjust to concurrent development of theory. Data collection was to continue until redundancy was achieved (Kuzel, 1992).

Data Sources

There were four proposed primary sources of information for data collection: DOII, SMQ-CF, NIH-CSCF, and interviews. The DOII and SMQ-CF were to be administered to each participant and data to complete the NIH-CSCF would be gathered by the investigator. Interviews were to be conducted based on selective and theoretical sampling from responses to the three instruments.

Demands of illness inventory.

The Demands of Illness Inventory was used to measure women's perceptions of the demands associated with CF on various aspects of their lives. The DOII is a 125 item instrument designed to measure thoughts and events experienced in response to health problems related to chronic illness (Haberman, Woods, & Packard, 1990). It contains 7 subscales: physical symptoms, personal meaning, family functioning, social relationships, self-image, monitoring symptoms, and treatment issues (see Appendix A). Items are scored on a 5-point Likert scale ranging from 0 to 4 (not at all to extremely). Higher DOII scores indicate higher demands of illness and higher ratings of intensity of demands. Cronbach's alpha for the entire scale has been reported at .96 (Haberman et al., 1990). Construct validity has been supported through both correlation with other standardized questionnaires and by the contrasted-groups method of validity. In this study, it was

proposed that participants respond using thoughts and events from the preceding 30 days. For this study, the subscale for physical symptoms was eliminated as measures were not relevant to CF (see Table 1). The SMQ-CF (described below) was used in its place to examine CF related physical symptoms. The final DOII inventory for this study consisted of 112 items. There is insufficient data to establish the reliability and validity of the revised instrument.

Self-management behavior form.

The research focus in this study was the women's ability to administer home therapy and their decision to seek treatment. The SMQ-CF measures a broad range of monitoring and therapy behaviors, many of which are not observable during clinic visits (Sockrider, Swank, Seilheimer, Bartholomew, Mariotto, & Parcel, 1996). An alternate instrument to measure self-management in CF was not known to exist. The SMQ-CF is a 46 item questionnaire that uses a 5 point response scale (see Appendix A). The scale was intended for use with CF caregivers and patients. Factors on the scale reportedly load according to 2 dimensions for a total of 12 factors. The two dimensions are content groupings (respiratory and gastrointestinal) and type of behavior (monitoring versus treatment) (Table 1). The 12 factors on the instrument include: observe digestion problems; observe other respiratory infection symptoms; observe chest physiotherapy and/or respiratory therapy; treat lower respiratory infection; observe nutrition; treat digestive/nutrition problems; keep treatment regimen; observe cough and sputum; adjust enzymes; take action to improve weight; not stopping treatment; and observe treatment of

respiratory infection. Internal consistency has been reported at .95 for the entire scale (Sockrider, Swank, Seilheimer, Bartholomew, Mariotto, & Parcel, 1996). The instrument has primarily been tested on caregivers of children birth through 18 years of age. It was used in this study with minor modifications.

The modifications included six additional questions. These questions were added to examine changes in disease management that were brought about by the pregnancy and by child rearing. The additional items examined transition as it related to personal meaning, symptom monitoring, and treatment issues. The revised instrument included 61 items. Reliability and validity were not established with instrument modification.

NIH clinical score for cystic fibrosis.

CF care consists of disease related interventions based on severity of illness obtained through physiologic measures. Physiologic measures such as pulmonary function tests (PFTs) and radiographic studies assist in determining severity of illness (Shwachman & Kulczycki, 1958; Sockrider, Swank, Seilheimer, & Schidlow, 1996; Taussig, Kattwinkel, Friedewald, & di Sant'Agnese, 1973). Fluctuations in the disease course of CF may be directly measured through these studies and may serve as a way to index disease status. Data from physiologic measures thus form a composite picture of severity of illness which could be used for comparison to self-report measures (Shwachman & Kulczycki, 1958; Sockrider, Swank, Seilheimer, & Schidlow, 1996; Taussig et al., 1973). While physiologic measures may be used to form a composite picture of severity of illness

for the purposes of comparisons, they have not been found to be reliable predictors of disease outcomes.

It was proposed that the NIH-CSCF would be used to obtain physiologic data (Taussig et al., 1973). These data were to be collected through chart review. The data to be collected included pulmonary function test data, body weight profile, activity, and attitude (Appendix A).

Completion of the NIH-CSCF results in an assigned numerical score that ranges up to 100; the higher the score the lower the severity of illness. The NIH-CSCF consists of 14 variables rated as point deductions with zero as normal. The total score is calculated by the subtraction from 100 of the sum of point deductions for 2 subscores: a) pulmonary (maximum deduction is 75 points) and b) general (maximum deduction is 25 points).

An internal consistency coefficient for the total NIH-CSCF of 0.81 has been reported (Sockrider, Swank, Seilheimer, & Schidlow, 1996). Items load on five factors: pulmonary, nutritional, disability, psychosocial, and acute infiltrate (see Table 1). The pulmonary subscore is reported to have an alpha value of .77. Data recorded under pulmonary are specific and detailed in regards to patient information. The general subscore alpha value is reported at .40. However, there are only three items-- weight, activity, and attitude--under the general subscore which may account for the variability; these did not correlate appreciably with total scores.

In this study, it was proposed the scale be used without modification. Data measurements were to be obtained for two to four points in time: at one month prior to

pregnancy, the end of the second and third trimester, and one month prior to participating in the study.

Pulmonary function test (PFT) data to be collected included forced vital capacity (FVC) and forced expiratory volume (FEV1). The FVC is the total volume exhaled (Voter & McBride, 1996; West, 1990). The FEV1 is the forced expiratory volume exhaled in 1 second. Normally the FEV1 is about 85% of FVC. The data provided from the PFT were to be used to identify and quantify the degree of obstructive disease present.

FVC and FEV1 are expressed as a percentage of predicted values (American Thoracic Society, 1991). FVC of predicted values are classified as follows: normal or >80%; mildly reduced or 65-80%; moderately reduced or 50-64%; severely obstructed or <50%; very severe <35%. FEV1 of predicted values are classified as follows: normal or >80%; mildly reduced or 65-80%; moderately reduced or 50-64%; severely obstructed or <50%; very severe <35%.

Certain genetic mutations of CF cause patients to have difficulties meeting nutritional requirements to maintain a desired weight of >90% ideal body weight. This is because of excessive caloric losses from malabsorption and additional energy demands of chronic endobronchial infections and increased work of breathing (Ramsey, Farrell, Pencharz, & the Consensus Committee, 1992). There is increasing evidence that nutrition and pulmonary status are closely linked. Males and females in all age groups with CF may be more poorly nourished than the general population in the United States (Rosenfeld et al., 1997). Body weight profile was the proposed measure for nutritional status. Body

weight profile is computed as a percentage of ideal weight for height and gender (percentage of ideal body weight) (Moore, Durie, & Forstner, 1985).

The final two areas on the NIH-CSCF are activity and attitude. These measure activity in relation to amount of energy to perform activities of daily living, exercise, and the ability to work or attend school. Attitude specifically addresses issues of compliance with care and depression. These two measures are based on the reports from health care providers and not the participants.

Interviews.

Open-ended and semi-structured participant interviews were proposed. Sampling for interviewing was to be based on scores from study instruments. The purpose of the interviews was expected to be provide more in depth analysis of the phenomenon of interest. Early interviews were expected to be broad-based, letting participants tell their stories. The proposed semi-structured interview outline is found in Appendix C. Subsequent interviews were to be focused on obtaining more targeted or detailed information as dictated by the emergent theoretical model. One interview per participant was planned; audio-taped interviews were to be transcribed verbatim. Reciprocal obligation to the women was to be met through scheduling of interviews at their convenience and thank you notes.

Table 1

Proposed Data Collection Forms

DOII: Sub-scales	**SMQ-CF: Factors**	**NIH-CSCF: Factors**
Cognitive functioning	Observe digestion problems	General pulmonary
Family functioning	Observe other respiratory infection symptoms	Nutrition (pulmonary)
Treatment issues	Observe CPT/RT	Disability* (activity)
Illness attribution	Treat lower respiratory infection	Psychosocial (attitude)
Emotions	Observe nutrition	Acute infiltrate
Body Image	Treat digestive/nutritional problems	
Symptom monitoring	Keep treatment regimen	
Personal meaning	Observe cough/sputum	
	Adjust enzymes	
	Take action to improve weight	
	Not stopping treatment	
	Observe treatment of respiratory infection	

* Defined- inability to perform daily activities such as work or school

Demographic Data

In addition to the DOII and SMQ-CF, it was proposed that demographic data be obtained from each participant. This information was to assist in case development for construction of data matrices or typologies. Demographic data for participants was to include date of birth, race, level of education, age at CF diagnosis, para/gravida, marital status, and employment. In addition, data on the children of the participants was to be collected including date of birth, sex, gestational age at birth, and sweat chloride and genetic testing results (if done).

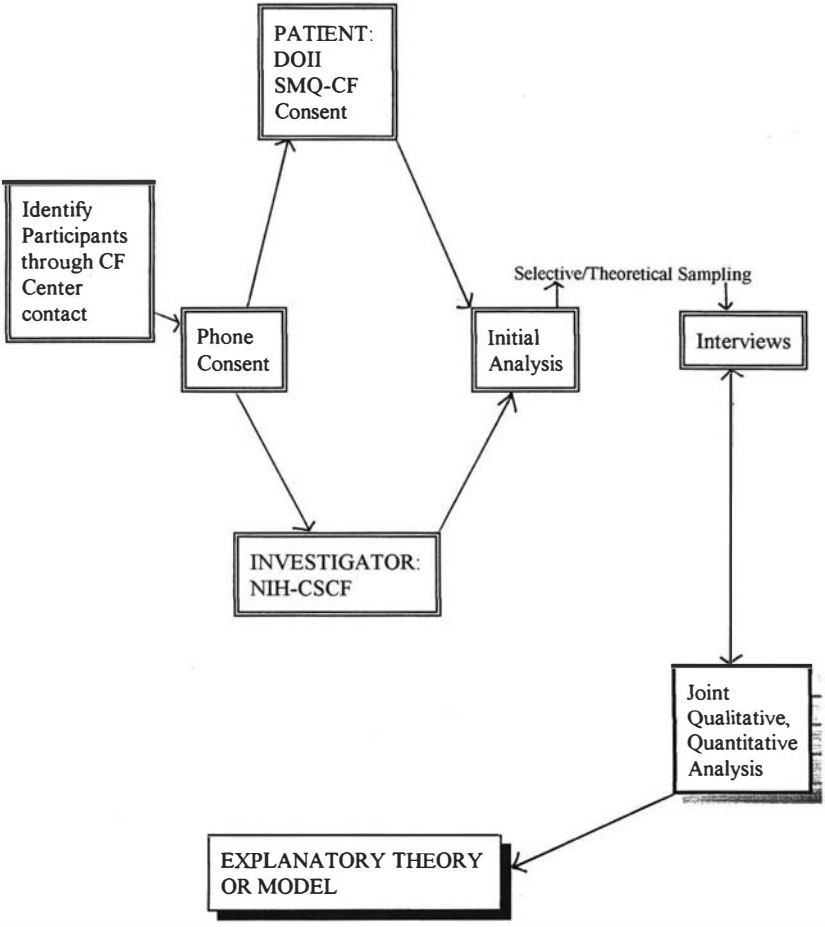
Procedure

Figure 1 illustrates the proposed process of data collection and analysis. Consent to conduct the study was to be obtained from each CF Center's Human Subjects Committee and medical director. A contact person was to be identified for each site. A letter of introduction, overview of the purpose of the study, length of time involved in participating in the study, and dates of data collection were to be mailed to each participating center's designated contact person. This person was to assist in identification and contact of eligible participants and in obtaining physiologic data for the NIH-CSCF.

Women eligible for the study were to be approached by the investigator either in person or by telephone to discuss the purpose of the study, answer questions, obtain phone consent, and arrange for delivery of the DOII and SMQ-CF. A research package

Figure 1

Proposed Flow of Data Collection and Analysis



with information inclusive of the DOII, SMQ-CF, and formal consent form to sign (with a copy for the participant to keep) was to be given or mailed to each participant (see Appendix D for consent). The informed consent form contained a request for permission to collect physiologic data from medical records and for a participant interview. A stamped self-addressed envelope was to be included with the research package for return to the investigator. A follow-up phone call was to be made if materials were not returned within 3 weeks.

Physiologic data for the NIH-CSCF was to be obtained from two sources. The first, the Epidemiologic Study of CF Patient Profile Report kept by CF centers, was expected to provide some data. The second source of information was to be the patient's medical record. The on-site contact person was to be requested to fill-out the NIH-CSCF using these data sources. The form completion was expected to take took ten to twenty minutes.

Data collection using the NIH-CSCF was to occur at two to four time points (depending on whether the woman was pregnant or had delivered her baby) and inclusive of: one month prior to pregnancy; the end of the second trimester of pregnancy; the end of the third trimester; and after pregnancy. The investigator was to make telephone calls and site visits as necessary to secure data.

The DOII, SMQ-CF, and NIH-CSCF were to be analyzed and the results placed in a data matrix. From preliminary analysis of the data matrix, selective and theoretical sampling would occur to begin participant interviews. The investigator proposed

recontacting women selected to for interviews. Consent was to be reconfirmed at the point of re-contact.

Problems Encountered During the Initial Phase of Data Collection

The original study proposal was approved on September 18, 1998. Initiation of the study protocol began October 1998. The initial proposal planned to recruit women directly through CF Centers. Three CF Centers had agreed to broker the study through their Human Subjects Committees in July and August 1998; three others had indicated an interest in October 1998. However, the proposal had not been introduced by December 1998 and follow-up with the Centers indicated a number of issues. The primary problem was in recruitment and the number of eligible participants. Recruitment efforts for the original dissertation proposal proceeded for approximately five months with one woman enrolled in the study from six sites by February 1999.

Despite assurances by CF centers prior to initiating the study, there were two reasons for the difficulties encountered with recruitment: inability of the investigator to penetrate CF center gatekeepers to conduct studies on CF patients, and a smaller number of eligible women than was originally calculated. Another problem contributing to recruitment was the extensive amount of research being conducted on the limited number of individuals with CF.

Consultation with dissertation committee members and external health care experts between December 1998 and March 1999 resulted in an alternative recruitment approach. The decision was made to contact women directly over the Internet and through an adult

CF newsletter. By appealing directly to women, CF Centers and the multiple barriers associated with them could be bypassed.

A research based web site was developed March 1999 and posted on the Internet and on two CF chat rooms. A research abstract describing the study in lay terminology was also placed in an adult CF newsletter (Appendix B). Interested women had the option to respond one of two ways, through the computer or over the telephone. Thus, study inclusion was not limited to only women with computer access.

The change in recruitment strategy dramatically changed study enrollment. However, while the recruitment rate improved, it was apparent that numbers would remain far less than originally proposed. The study was now one year post proposal defense and included eight women. Discussions with the dissertation chair and committee members supported a change in sample size and methods from the original proposal.

A second, complicating factor emerged from preliminary data analysis. What emerged from demographic data was a sample that was far different from what was described in published literature and from that which was described in the dissertation proposal. Nearly half of the women who had responded to study advertisements had been diagnosed with CF after the age of 18 years. Moreover, some participants had been diagnosed after the birth of their children.

Summary

Based on the small sample size and unusual nature of the demographics of the women who responded to recruitments efforts, the dissertation proposal shifted focus

from a quantitative approach with a small qualitative component, to a project that was predominantly qualitative with a small quantitative aspect. Research techniques changed with primary emphasis shifting from research questionnaires to the participant interview. The interview, while partially structured by the individual's responses to research instruments, also followed a semi-structured interview outline. A detailed description of the actual methods used in the study is found in chapter four.

Chapter 4

Revised Research Design and Methodology

The difficulties encountered with recruitment of subjects and the paucity of women available who were eligible for the study, necessitated changes in the originally proposed methods. The discussion presented in chapter four describes the actual methods used in the study.

Design

The design of the study remained grounded theory method, as interpreted by Glaser (1978). However, the focus of the study shifted to the examination of women's perceptions of how the demands of having a chronic illness (CF) influenced their ability to care for their children and continue to meet their own health care needs.

Two forms of data collection techniques were employed; one was quantitative for use with research questionnaires and the second was qualitative for participant interviews. To accommodate two different but complementary research techniques, a complementarity model was used to guide analysis. A complementarity model was indicated because the study used qualitative (words) and quantitative (numbers) to measure overlapping, but distinct facets of the phenomenon under investigation (Caracelli & Greene, 1993). This model permitted the use of results from one data collection technique to elaborate or enhance the results from another (Caracelli & Greene, 1993; Greene, Caracelli, & Graham, 1989; Morgan, 1998). For this study, data collection was

initiated with quantitative instruments for the purpose of situating or guiding the women and interviews with the investigator following receipt of study instruments.

The goal in combining data sources was to achieve fuller description, not convergence. The phenomenon of interest had a limited range and the use of a combined or mixed method permitted examination of different facets of it. The expected outcome remained a theoretical model to elucidate relationships between disease and child care demands in women with CF.

Sample

The population of interest were women with CF who had given birth to one or more living children. The technique for sampling was changed from recruitment through CF Centers. The sample was recruited primarily from two sources: the sources were the Internet with postings on two CF chat rooms and a research Web site, and through an adult CF newsletter (Appendix B). The research web site was posted in March 1999 and remained in place for 12 months. The advertisement in the adult CF newsletter occurred three months after chat room postings. The purpose of spacing recruitment efforts was to stagger participants out over a period of time to permit full analysis of data.

Women included in the study met the following inclusion criteria: eighteen years or older, had a diagnosis of CF, and had a living child. All women who met sampling criteria were included in the study. Final sample size was based on the number of women who responded to advertisements and on the number of women required for analysis in order to develop a conceptual model around the phenomenon of interest.

Data sources.

The four primary sources for data collection remained the DOII, SMQ-CF, NIH-CSCF, and participant interviews. The DOII, SMQ-CF, NIH-CSCF, and participant interviews were collected concurrently on each woman (Appendeces A and C). These were described in detail in chapter three.

Demographic data.

In addition to the DOII and SMQ-CF, demographic data was obtained on each woman. Demographic data for participants included date of birth, race, level of education, age at initial CF diagnosis, para/gravida, religious preference, marital status (or relationships), employment, and insurance status. Data on children included date of birth, sex, gestational age at birth, and sweat chloride and genetic testing results (if done).

Procedure

Consent to conduct the study was obtained through the Human Subjects Committee from the originating university. The data collection procedure included a combination of techniques and tools to elicit information concerning the phenomenon of interest. Data collection was done concurrently. A specific sequence of events occurred in the process. Figure 2 illustrates the revised process of data collection and analysis. The next series of procedural steps related to recruitment, data collection, and initiation of analysis.

Recruitment.

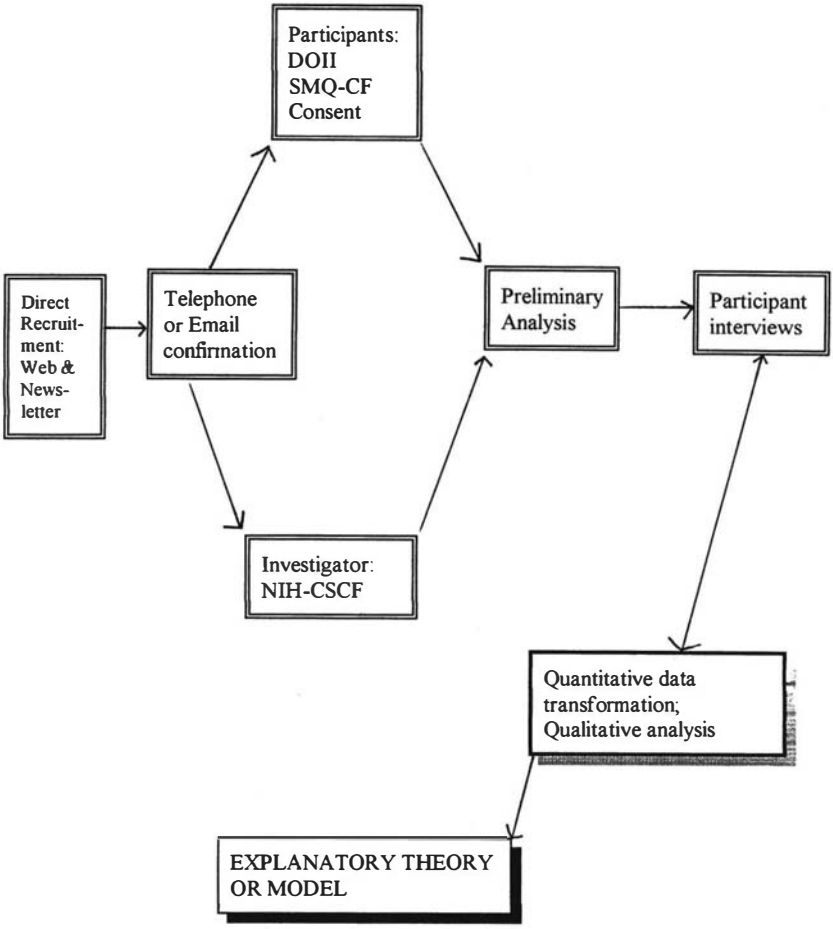
Several strategies were used to recruit women for the study. One mechanism was the development of a research web site. The web site contained a description of the study, autobiographical information about the researcher, eligibility required to participate in the study, additional web sites to contact for general CF information, and the investigator's Email contact address for women interested in participating in the study. Web site information was posted in an adult CF newsletter and on two CF focused chat rooms (Appendix B). The adult newsletter also included a telephone number for women who did not have access to a computer to contact the investigator.

Women indicated their interest in participating in the study either through the telephone or computer Email. Following contact by the women, the investigator followed-up either through Email or by telephone to discuss the purpose of the study and the length of time involved in participating in the study, answer questions, and arrange for mailing study instruments and consent forms. Because of an inability to guarantee confidentiality over the Internet, the package of research materials was mailed through the postal service (Sharf, 1999; Sudweeks & Simoff, 1999).

All women who agreed to participate in the study were asked to complete a mailed packet of information. A stamped, investigator-addressed envelope was included in the packet in which to mail forms back to the investigator.

Figure 2

Actual Flow of Data Collection and Analysis



Data collection.

The research packet mailed to each woman included a letter to follow-up confirmation of participation and an explanation of the enclosed materials, two copies of the informed consent form (one to keep and the other to return), the Demands of Illness Inventory (DOII), the Self-Management Behavior Form (SMQ-CF), the demographic form, and a stamped investigator-addressed envelope in which to return materials. Follow-up was done with participants either through Email or the telephone if materials were not returned within three weeks.

The mailed research package also included a form for the women to identify the name, address, and telephone number of the site where CF care was received. Each woman was asked to identify a contact person at this site in order to receive medical records information for the NIH-CSCF. The contact person (health care provider or CF Center) was mailed the NIH-CSCF along with a copy of the signed consent form granting permission to obtain medical records information. Center contacts completed the NIH-CSCF. The investigator made follow-up telephone calls to the on-site contact person to answer questions.

The participant interview was the final component in the sequence of data collection. An appointment was scheduled with each participant to set up a convenient time when she could be undisturbed by work or child care responsibilities. All interviews were scheduled for a one hour time period and were recorded by hand. Interviews were

conducted after receipt of the DOII and SMQ-CF. However, receipt of the NIH-CSCF by the investigator was not necessary to conduct interviews.

The DOII and SMQ-CF also served as an elicitation device for participants prior to being interviewed. It provided participants with information (mediated through the use of the questionnaires) in preparing for interviews to trigger more thoughts about the phenomenon of interest. Interviews also served to clarify thoughts or events that occurred during the time when questionnaires were being completed. Participant's responses to each part of the research had the potential of being affected by disease exacerbation or recovery. For example, if participants were entering an exacerbation or were recovering from one, their responses on the questionnaires might be reflective of those transition events in their lives.

Initiation of analysis.

Following receipt of the DOII and SMQ-CF, a data matrix was developed for each participant arranging each item on the DOII from highest to lowest intensity of demand. Information from the SMQ-CF was arranged based on intensity of care, responses to specific health care needs, and a list of daily treatments. Data matrices ultimately contained NIH-CSCF scores and themes from analysis of participant interviews.

There were two objectives for arranging data in a comprehensive table or data matrix. The primary purpose was to organize data for within and between case analysis. Within case analysis assisted in identifying themes or patterns for each participant.

Secondly, from preliminary within case analysis of the DOII and SMQ-CF, questions were organized to frame participant interviews.

Analysis of Data

The constant comparative method associated with grounded theory method served as the basis for analysis (Glaser & Strauss, 1967; Strauss & Corbin, 1994). As a result of this interactive process, data analysis occurred at several levels. These levels were research questionnaires, participant interviews, and within case and between case analysis.

Combining Methods–Integrative Analysis Strategy

Data from the DOII, SMQ-CF, NIH-CSCF, and interviews were placed in a data matrix for analysis as a single case for each participant. Analysis of raw data began with receipt of the DOII and SMQ-CF and the development of a data matrix for each participant. The data matrix was examined and analyzed looking for patterns in participant responses. The patterns that emerged from analysis of each participant's responses on the DOII and SMQ-CF served to help prepare the investigator for interviews based on participant responses. Within case data entry and analysis continued with the receipt of each participant's responses to the research instrument being entered into the matrix (e.g.–analysis from participant interviews, NIH-CSCF). In this way, there was an ongoing comparative interaction between data collection and analysis.

Analysis progressed from within case to between case comparison and analysis as patterns of responses continued to be elucidated. Between case analysis compared patterns that emerged from within case (each participant) and with further abstraction of data, then

went on to form categories. With each step in analysis, increased abstraction of data permitted the move from description, to analysis, and finally, synthesis and interpretation.

Comparisons of the outcome of data analysis were made with the theoretical framework outlined in the proposal. The anticipated outcome of data analysis and comparison with the proposed theoretical framework was improvement of the existing theoretical model with the phenomenon of interest or towards conceptualization and early theory development to fill perceived gaps.

DOII, SMQ-CF, and NIH-CSCF.

The DOII, SMQ-CF, and NIH-CSCF were analyzed two ways. Data from the questionnaires was placed into a data matrix for each participant. These matrices continued to develop throughout the research process until they contained information from all sources of data collected on the participants. The second form of analysis was through descriptive means.

The first step with research questionnaires was to set up data matrices. Responses to research instruments were placed into data matrices, with one for each participant. The data displays that were portrayed in the matrices permitted visualization of response patterns of the participants for the purpose of within and across case comparisons.

Items from the DOII were arranged from highest to lowest intensity of demand. The SMQ-CF listed intensity of home care required according to each affected system (e.g.- respiratory–chest physiotherapy (CPT) four times per day). The NIH-CSCF score was reported as a single number for each factor of pulmonary, attitude, and activity.

Research questionnaire measures were also examined through descriptive means. The data was entered into SPSS 7.1. However, the sample size was too small to attempt any meaningful group statistical analysis. Each research instrument grouping entered into SPSS 7.1 was then viewed as a single homogenous grouping independent of the other instruments (e.g.--all DOIIs without regard to participant and homogenous grouping of scores on the NIH-CSCF or SMQ-CF).

The DOII items on the questionnaires were visually examined and compared for frequency and intensity of demands of illness. In other words, the DOII responses were examined for the most frequently occurring, highest demand item numbers. The SMQ-CF was viewed for type, amount, and intensity of daily therapies as well as reported frequency of missed therapies and management of disease symptoms of each participant score against others. Lastly, the NIH-CSCF was reviewed for scoring of factors within pulmonary, attitude, and activity. Of interest were any patterns or themes that might have emerged from comparison of these homogenous data groupings.

Interviews.

Participant interviews occurred concurrently during data collection. There were two ways in which this was planned and integrated into the research. First, participant interviews were scheduled within thirty days of receipt of completed research questionnaires. Secondly, hand written interview notes were immediately transcribed and analyzed. This permitted ongoing comparison and refinement of emerging patterns and themes to the phenomenon of interest.

Content analysis of interviews occurred immediately following the formal telephone interview. Transcriptions of interviews underwent a process of data deconstruction and reconstruction. The raw data from transcribed participant interviews was deconstructed into data units. The thoughts, responses, and terms used during interviews by participants were organized into data units. Data units contained groups of related thoughts or ideas shared by the participants. Data units were compared to every other data unit processed for data analysis for further data reduction through comparative pattern analysis.

Comparative pattern analysis assisted data units to be processed into categories. Through elucidation of key phrases or terms from interview transcripts, the developing category system remained close to participant responses about the phenomenon of interest. In other words, the language was that of the participants and not the investigator. Developing categories were evaluated for internal homogeneity and external heterogeneity. In this way, categories emerged that contained data units related to the same content and were not redundant. Data reduction continued with combining homogeneous data units under different categories. Additional data was sought from each participant subsequently enrolled in the study, to provide depth, understanding, and confirmation to emerging categories.

As categories became more saturated, they were compared to one another in an effort to identify dominant concepts. The purpose of comparison of categories was to begin to analytically connect and integrate data. During this phase of analysis, data

reconciliation or reconstruction was begun. Reconstruction or the reconciliation of data was begun in the form of weaving together influences resulting from the categories which contained individual data units. This interweaving was accomplished by comparing emerging concepts and their properties to each other to begin theoretical sorting. Sorting of ideas permitted integration of concepts into existing or emerging theory.

The final level of analysis was sorting and synthesis of emergent concepts to determine how it would fit into the theoretical framework of transition theory proposed in the study. The emerging concepts were analyzed for commonalities and differences within and between the categories of data sets. This analysis was used to begin to develop and test propositions either add to transition theory or to begin to construct an alternate explanatory theoretical model. Preliminary testing of the propositions occurred through interviews with the last four participants. Questions were added to participant interviews related to these propositions.

Strengths and Weaknesses of the Study

The strength of the study was in the depth that a multi method approach to the phenomenon of interest permitted. This approach allowed for discovery and voice to be given to the participants. The descriptions of the categories in the theoretical model that was developed are those of the participants.

The problems that were encountered at the onset of data collection represent the limitations of what is known about this group of women. One of the initial criticisms of the

few studies that have been conducted is the small sample size. Despite a variety of techniques to attract potential participants, this study was also limited in its sample.

The emphasis of the focus and the context of the study is representative of only the experiences of the participants. It also represents the experiences of participants with computer and Internet access and skills. The study is not intended to be generalized to other mothers with CF.

Summary

The move from examining data matrices as an entity, looking at patterns, associations, causes, and effects within cases, to comparative analysis between cases, condensed data into a more abstract form. The analytic progression moved from telling individual stories about a specific phenomenon to constructing a "map" which contained more formal elements of the "story" and identified key concepts (Miles & Huberman, 1994; Sandelowski, Holditch-Davis, & Harris, 1992). The final step of analysis served to further synthesize and interpret data in order to improve existing theories and to begin to develop an explanatory model.

Revised Research Questions

The initial question that was proposed at the onset of this research was how the disease of CF was impacted by pregnancy and child rearing. A smaller than projected sample size of participants could not answer this question of change in illness severity. Additionally, the relatively healthy physical status of participants did not support a disease focused approach to the question.

Rather, the revised methods used in the study resulted in a re-focus of the study. The revised aim was to understand how women with CF incorporated caring for a child with continuing to care for themselves and their disease. The research questions also evolved to focus on the three primary influences on women's daily lives: impact on health, demands, and supports. The revised research questions were:

1. How did women with CF view the impact pregnancy and caring for children had on their health?
2. What demands were placed on women in caring for their children and their CF?
3. What supports did women with CF perceive as needed to care for their children and manage their CF?

Chapter 5

Research Findings

The purpose of this study was to examine what demands chronic illness placed on mothers with CF. The specific aim was to understand how women with CF incorporated caring for children in addition to their disease and its treatments. Three areas were explored: impact on disease, demands of illness, and supports. The following research questions were addressed:

1. How did women with CF view the impact that pregnancy and caring for children had on their health?
2. What demands were placed on women in caring for their children and their CF?
3. What supports did women with CF perceive as needed to care for their children and manage their disease?

Sample Characteristics

Sixteen women responded to recruitment announcements. Women were provided with the opportunity to request participation in the study through the use of the computer or telephone. Eight participants (50%) were recruited over the Internet, seven (44%) to an announcement placed in an adult CF newsletter, and one was obtained through requests to six CF Centers. Fourteen of the women who expressed an interest in participating in the study did so via Email and one participant contacted the investigator through the telephone. Seventy-five percent of respondents completed the study: eleven participants

completed both research instruments and interviews and one participant completed only an interview (n=12).

Characteristics of Participants

The age range for participants was 18-57 years. The range in age for first pregnancy was 17-33 years. The participants were all Caucasian. The age at diagnosis for CF ranged from newborn to 36 years.

Two distinct groups of women emerged from within the sample who responded to the three modes of recruitment. Women in group 1 were diagnosed with CF in childhood (n=7). The women in group 2 were diagnosed after the age of 18 years, and in two cases after pregnancy (n=5). CF diagnosis in Group 2 was reported by the women as having occurred generally in response to perplexing chronic respiratory symptoms, most notably severe sinus disease. Despite the lack of a childhood diagnosis, women in Group 2 reported disease symptoms similar to the women in Group 1. Four out of five participants in Group 2 described themselves as being too ill to be employed compared to four out of seven participants in Group 1. Table 2 contains summary data for both groups of participants, describing age at diagnosis, age at first pregnancy, and present age at the time of participation in the study.

Manifestations of CF symptoms.

The participants had a broad range of CF symptoms and were treated with a variety of therapies. As part of the study, participants were asked to complete the SMQ-CF questionnaire outlining symptoms, daily therapies, and decision making strategies

Table 2

Characteristics of Participants

GROUP 1			GROUP 2		
Age at Diagnosis	**Age At First Pregnancy**	**Present Age**	**Age At Diagnosis**	**Age At First Pregnancy**	**Present Age**
4 years	23 years	25 years	18 years	18 years	32 years
1	26	34	32	33	36
11	19	57	19	25	39
4	28	31	36	19	37
3	27	32	33	28	34
Birth	27	35			
Birth	17	18			

when confronted with specific changes in CF symptoms. Reported symptoms and manifestations of CF included but were not limited to sinusitis, infertility, mild to severe respiratory symptoms, and double lung transplant. Table 3 provides an example of how one section of the SMQ-CF was analyzed from one participant.

All participants were on some form of routine therapy including one or more combinations of the following: daily inhalation treatments, chest physiotherapy, pancreatic enzymes, and antibiotics. None of the participants reported being routinely on oxygen therapy at the time they participated in the study.

Table 3

Sample Analysis of One Section for One Participant's SMQ-CF

Think about how you treat a lower respiratory infection. **Circle** how frequently you do each action. Note: If you have not had a lower respiratory infection (bronchitis/pneumonia), **skip** to question **140.**

	0=not prescribed 1=Never 2=Rarely 3=Sometimes 4=Usually 5=Always					
132. Begin respiratory therapy (aerosol, inhalation) or increase the number of respiratory treatments done each day?	0	1	2	3	**4**	5
133. Begin chest physical therapy (CPT) or increase the number of CPT sessions done each day?	0	1	2	3	**4**	5
134. Begin prescribed antibiotics or increase number of antibiotics used with the advice of your doctor's office?	0	1	2	3	4	**5**
135. Arrange daily time schedule to fit in extra therapy?	0	1	2	**3**	4	5
136. Contact the doctor's office if no improvement occurs after the change in therapy?	0	1	2	**3**	4	5
137. Continue treatment at the end of a prescribed course if the symptoms have not disappeared?	0	1	2	3	4	**5**

Average Score for the subsection Treat Respiratory = 4.7
(Bold and underlined numbers indicate the participant's responses on the questionnaire)

The participants' health care providers were requested to complete the NIH-CSCF, which provided a numerical score based on the cumulative health of each woman. This score was included in the study to provide information about PFT's and potential

complications associated with CF. The health information for the NIH-CSCF was obtained from participants' medical records and permitted a more longitudinal view of their CF. For example, cor pulmonale, certain x-ray findings, pulmonary surgery, and the number of pulmonary exacerbations are all evidence of severity of illness indicators.

The NIH-CSCF scores from the women who participated in the study ranged from 46-81 (possible maximum score of up to 100). Higher scores indicated better health. Comments provided by the health care providers indicated that most participants' scores had remained relatively unchanged during and following pregnancy. Appendix A provides the study form as a point of reference.

Lastly, three participants reported significant changes in lung microbiology with acquisition of multi resistant bacteria. The changes contributed to enough of a decline in health to allow two of the participants to become eligible for disability and for the third to ultimately require a double lung transplant as a consequence of the severity of decline in her pulmonary function. Changes in pulmonary function for these participants were reported both by telephone and in handwritten notes by health care providers as a consequence of the disease of CF and were not thought to be related to child bearing or child rearing.

Knowledge of pregnancy.

Awareness of fertility and pregnancy in women with CF has until the past decade been more a question of debate than reality. With the increase in age of survival, more women have become pregnant. Seventy percent of the women who participated in this

study reported they were aware they could get pregnant. However, it was not something that was routinely discussed as part of CF visits nor included with education surrounding their CF diagnosis. Consequently, 55% of participants reported unplanned pregnancies.

Education and employment.

The challenge of having a chronic illness did not deter many of the participants from pursuing college and/or working. All but one participant had completed high school. Fifty-eight percent of the participants had completed college degrees and 8% had pursued graduate education. Forty percent of the participants were able to continue working twenty hours or more per week as well as care for themselves and a family. Sixty percent of the participants were on disability or not employed because of the severity of CF symptoms. One participant reported being on disability but also continued to work from her home.

Characteristics of partners.

The women who chose to participate in the study were requested to provide information on their partners and their decision to undergo CF testing. Two of the fathers of the children underwent testing for CF. One father had negative genetic testing; the second father had a gray or borderline abnormal sweat test and was undergoing further studies. The other participants indicated that their partners were not interested being tested for CF because of the perception that there was a low probability of their being carrier.

Characteristics of the children.

The twelve participants had a total of twenty children. Table 4 contains demographic information on the participants' children. The participants and their partners were provided with the option of having their children tested for CF. CF testing could being done during pregnancy or after birth of the child. Only one of the participants had chosen to have an amniocentesis with CF testing (which was negative). The other participants did not choose testing during pregnancy as they were not planning to abort a fetus with abnormal genetic results.

CF testing after birth occurred for fourteen of the twenty children. Ten percent of participants chose not to have any form of CF testing done on their children. Seventy percent of the children of the remaining participants who had CF testing had a sweat test and twenty percent had genetic testing (one child had both studies done). Test results were variable for CF. Two children had positive sweat tests, one child had a gray or borderline abnormal sweat test, and one child had a positive genetic test.

Participant Data Matrices

Participant data matrices contained information for every data point recorded. The participant data matrices with complete data were joined together to form large tables for manual examination. Table 5 provides an example of a data matrix developed for the purposes of analysis. In an effort to protect participant confidentiality, only selected segments of information are found in this table.

Table 4

Characteristics of the Children

AGE	SWEAT TEST	GENETIC TESTING	AMNIOCENTESIS
2 years	negative		
8 years			negative
30 years	negative	negative	
35 years	negative	negative	
38 years	negative	negative	
6 years	gray (borderline)		
10 years	positive		
13 years	positive	positive	
14 years	negative		
3 years	negative		
3 years			
9 years	negative		
4 ½ years	negative		
14 months	negative		
8 years	negative		
12 years			
20 years			
3 years			
6 years			
1 month	negative		

The factors from study forms to which participants responded are found in Table 1. Study forms with numbered items and scales are in Appendix A. Table 5 is set up in columnar format with a column for each questionnaire. Outlined under the subtitles are samples of data from participant data matrices. For example, under the first column, DOII, the item number refers to the statement to which the participant responded. The items are listed under the level of intensity reported by the participant.

Each participant had a series of data matrices which contained data that over time became condensed into common patterns with fewer data points. The way in which analysis proceeded was to set up the first data matrix. Secondly, each column in the matrix was manually examined for common patterns of response. These common patterns of response were recorded in a second blank matrix that was set up identical to the original. A third matrix was formed from data from the second matrix. The patterns were arranged into themes which were next examined to form categories that were unique and did not overlap. This comparative process continued throughout data analysis. As a result of the ongoing comparative process and condensing of data, columns of data ultimately collapsed into a solitary list of categories with defining characteristics for each participant. Table 6 illustrates an example of the process for two categories for one participant.

The next level of data analysis compared categories across cases (participants). Categories across cases were examined for differences and commonalities and ultimately merged to form a single set of categories with defining characteristics for the entire group

Table 5

Example of a Participant Data Matrix Developed for Within and Across Case Analysis

DOII Item #	DOII Subscales	SMQ-CF	NIH-CSCF	Interview
Level 4 intensity	Level 4	Factors	Factors	Themes
■ 9	Cognitive Functioning	Observe respiratory	Gen. Pulmonary-	Pregnancy-
■ 109	Symptom Monitoring	● Mean	● 46	-aware/planned -"fixated" on health and mortality
Level 3 intensity	Level 3	score=4.8	X-ray = 13 PFTs = 15 Exacerbation = 5 Hemoptysis = 3 Pulmonary Surg. = 0 Cor pulmonale = 3 PE = 5 Sputum = 2	- a > awareness of need to stay healthy/do care
• 1	Treatment Issues	Observe CPT		Fatigue -
• 2	Family Functioning	● Score = 2.8		-prioritize event
Level 2 intensity	Level 2	Treat respiratory		-how to fit in CF with care/child/home/work
▸ 4	Emotions	● Score = 4.8		Finance - insurance issues
▸ 12	Symptom Monitoring	Observe digestive	Nutritional	
Level 1 intensity	Level 1	✓ NA	✓ 0	Supports - family, friends, and work with health care providers
♦ 3	Body Image	Treat digestive	Disability/Psychosocial	
♦ 7	Level 0	✓ score = 5.0	▸ 5	
Level 0 intensity	Illness Attribution	Keep treatment Rx	Total Score	
		■ Score = 4.5	*100-51 = 49	

of participants and may be found in Table 7. The theoretical model depicted in Figure 3 was developed through this analytical process.

The next section of this chapter discusses this preliminary theoretical model that ultimately emerged through data analysis. The discussion is subdivided into three areas. The first area will discuss the fit of transition theory to the theoretical model that emerged

Table 6

Example of Patterns, Themes, and Categories for One Participant

Patterns	Themes	Categories
1. A housekeeper gives me more time with my children and time for treatments. 2. When my husband hears my coughing more, sleeping more, or observes other changes in my CF, he immediately takes over my responsibilities around the house so I can take care of my symptoms before they get worse and I need to be hospitalized. 3. We decided to hire a lawn service so he could help me out more with our children and around the house. 4. If you can afford to have help it really makes a difference in doing what you need to do for your self and not worrying about how to do it all.	1. What is helpful in order to get housework done. 2. What is helpful to free up time for the family to do what is needed. 3. What is helpful to give the participant time to do her CF care. 4. What is helpful to the participant when she has an exacerbation. 5. Learning how to ask for and use help to care for self and children.	Home Support Defining Characteristics: 1. Things that enabled her to take care of herself and her family. 2. Use of people to help maintain the household (division of labor).

Patterns	Themes	Categories
1. The Internet gave me the opportunity to read more about the latest in CF therapy. I use it at least weekly. 2. My doctor never shared any information about CF and being pregnant. I had to find other ways to obtain information. The CF team seemed angry when I tried to discuss things I learned. I got so frustrated I finally changed doctors. I have to drive much farther but at least they work with me not against me. 3. I found it useful to use the chat rooms to talk to other women with CF who had been pregnant. They directed me to other resources I would not have known had we not had a computer.	1. How the Internet is used by the participant. 2. Information sharing by the health care team. 3. What the participant felt was important to know about being pregnant and having CF. 4. How the participant went about getting the information she thought she needed. 5. Learning how to ask questions and follow through to get an answer.	Obtaining and communicating information Defining Characteristics: 1. Use of the Internet and other resources. 2. Information about being pregnant and having CF. 3. Knowledge and information needed to care for self (CF).

from data analysis. The second area will present story sequences that serve to illustrate the theoretical model. The final area will explicate answers to the research questions.

Theoretical Model

The theoretical framework for this study was transition theory. The framework functioned as an organizing framework to initiate analysis of data. The interpretive

Table 7

Categories and Defining Characteristics

Category	Defining Characteristics
Knowledge	1. Information needed to deal with disease and treatments. 2. Information about pregnancy and having CF. 3.Use of the Internet/other resources.
Financial	1. How to finance maintenance needs of CF. 2. How to finance illness. 3. Personal finances and family needs. 4. Disability versus work issues. 5. Adequacy of health insurance. 6. Ability to use of CF knowledgeable physicians for health care.
Level of Energy	1. Role of fatigue in care for self and children. 2. Consequences and toll on social and personal activities.
Home Support	1. Those things that enabled participants to keep house, care for the family, and care for themselves. 2. How/who cleans house, laundry/dry cleaning, and outside work on house and yard. 3. Reliable child care. 4. The assistance that is needed to free up time for taking care of their health. 5. Learning how to ask for help.
Health Care Team Support	1. Relationship with health care providers. 2. Support provided by health care team. 3. Extent to which health care team works to integrate the participant in the plan of care.

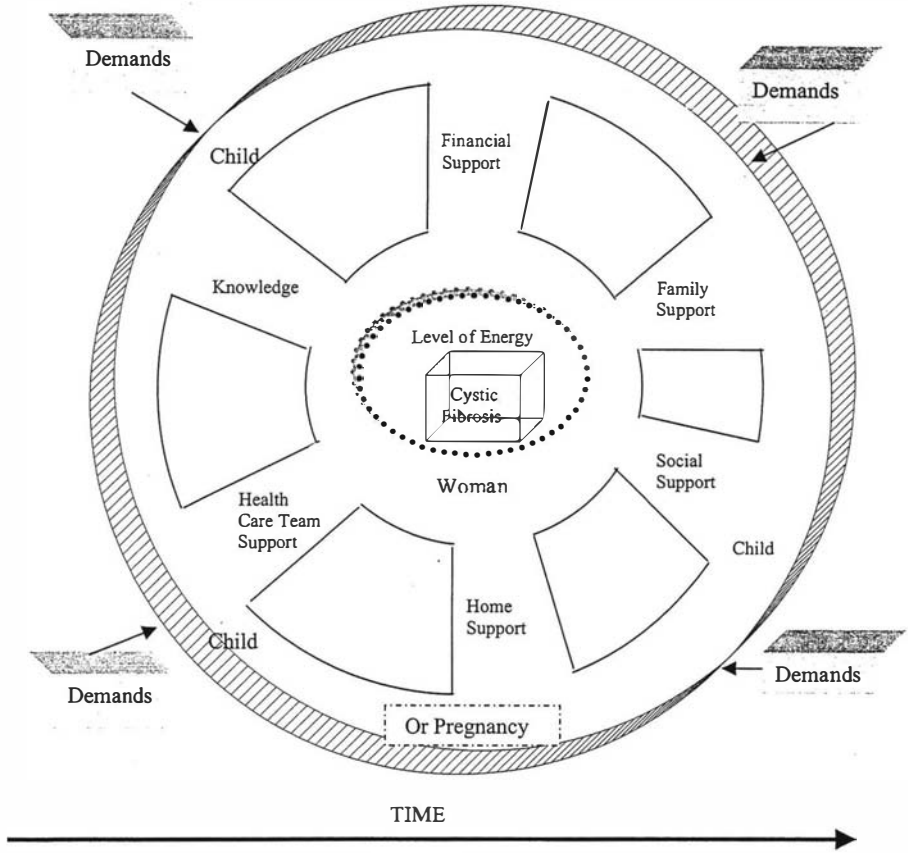
Category	Defining Characteristics
Family Support	1. Support of partner or spouse. 2. Support provided by nuclear and extended family.
Demands	1. Transitions. 2. Growth and development. 3. Level of energy. 4. Physiology of CF disease.
Social Support	1. Support of friends, community, and work. 2. Ability to engage in social activities for self and family.

product that emerged was grounded in the participants' responses to research instruments and interviews.

The specific aim of this grounded theory method study was to develop an understanding of how women with CF managed the demands of their children and their disease. Research data from questionnaires and interviews were used to develop a preliminary theoretical model of this phenomenon. Figure 3 depicts the theoretical model that was developed. The theoretical model entitled Elements Necessary to be a Mother with CF, reflected social, medical, and economic themes of how participants worked to sustain and maintain family life and a chronic illness entering the twenty first century. The model is compared for fit with transition theory in the following section.

Figure 3

Elements Necessary to be A Mother with Cystic Fibrosis



Demands

Transitions
Growth/Development
Level of Energy
Physiology of CF Disease = (CF symptoms, CF exacerbation)

The fit with transition theory.

Transition theory, as initially conceptualized for this study, was re-examined to determine the logical adequacy and usefulness to the phenomenon under investigation. Transitions that were theorized to be associated with CF were developmental, situational, and health-illness. Of interest was the relevance of transition theory to the descriptions of participants. The patterns of response that emerged through data analysis were examined for their relationship to this transition framework.

One participant commented during her interview that "CF doesn't run my world...it's always there". In examining the "fit" of transition theory to the experiences shared by the participants, transition theory captured some of what was described. Transition theory was appropriate for some of the developmental, situational, and health-illness issues. For example, events including birth, divorce, new diagnosis, and transitioning from work to disability, clearly encompassed transition theory and helped in understanding the response and meaning of changes occurring in the participant's "assumptive world". However, participants' responses reflected both short and long term struggles of having a terminal chronic illness and maintaining a family. The struggles the participants' described were ongoing–there was never a beginning nor an end as proposed by transition theory. What was problematic was understanding how the constancy of CF, "its always there", impacted participants' responses to and ability to manage their disease and children.

Transition theory was only part of the theoretical framework needed to develop an understanding of the phenomenon being described by the participants. What emerged from analysis of data revealed several other important components. The components of the model are what emerged as categories from data analysis: social support, financial, knowledge, level of energy, family support, health care team support, demands, and home support. These eight categories included both the demands as well as supports needed to deal with the demands. The categories that became the components of the theoretical model were described by the participants as crucial in learning how to successfully manage their disease and be a mother.

The theoretical model that was developed from the research.

The patterns, themes, and categories with defining characteristics that emerged from the empirical data were re-examined and a preliminary visual framework was developed to represent the phenomenon from the participant's point of view. The model, a preliminary view of the phenomenon, is descriptive. In attempting to understand how participants integrated caring for children and CF, the model was developed incorporating participant descriptions solicited from interviews. In this way, the model was grounded in the data. What follows is a description of the model, Elements Necessary to be a Mother with CF. Specific participant examples are provided in the section on story sequences.

The descriptive theoretical model, Elements Necessary to be a Mother with CF, (Figure 3) was visually depicted as a series of circles and abstract shapes. The inner most shape represents CF. The participants were clear in their interviews that while CF was at

the center of their lives, it did not dominate the their lives. CF was always "there" but it did not define them. Rather, the participants felt they were defined by other factors in their lives.

The next shape represents level of energy. Level of energy was described by the participants as constantly rising and falling. When energy levels were high, they were reported as a means of support. Conversely, when levels of energy were low they became a demand on participants. As a result of fluctuating levels of energy, participants reported planning activities based around how they felt. When energy levels were approaching their lowest point, participants reported seeking medical intervention. The more intense dips frequently represented one of the symptoms of a CF exacerbation.

The third shape represents the participant. This circle is influenced internally by the CF, but is not always dominated by it. The participants were also influenced externally. These external factors are represented by the six spokes that surround the second circle. The six spokes are analogous to a wagon wheel. The spokes represent the categories that emerged from descriptions by the participants as crucial in enabling them to be successful in caring for themselves and their children. When fewer of these spokes were present (if one of the categories was removed or absent), participants described increasing difficulty and problems in caring for themselves and their children.

The eight categories that were described by the participants included both demands and supports. The supports included knowledge, financial, home support, health support, family support, level of energy, and social support. The demands were the demands and

level of energy. Level of energy fluctuated between being a demand and a support depending on how the participant was feeling. The categories and their defining characteristics are found in Table 7.

The final circle represents the child and/or pregnancy. All the participants described the critical importance of the role these two experiences (being pregnant and having a child) assumed in their lives. As a result, metaphorically speaking, when spokes in the theoretical model of the wheel were missing or broken, participants described placing the needs of their children before their own in an effort to maintain balance.

The spoked wheel is surrounded externally by arrows that represent the demands that participants encountered. The responses reported by participants to these demands were influenced by the strength of the infrastructure of the wheel. The response was measured by the report of intensity to the DOII as well as through participant interviews. The more supports that were reported intact or in place, the less intense the demands.

There was one important over riding influence on the model–that of time. Time seemed to be primary to all situations described by the participants. In the model, it is depicted as a one directional arrow because time cannot be reversed. Listed under time are the demands that are also time oriented and time driven. These are transitions, growth and development, level of energy, and the physiology of CF.

The defining characteristics listed under the one directional time line arrow were the demands on the participants when they experienced a change in one of these areas (i.e.- pulmonary exacerbation, divorce, loss of ability to work because of CF). The

intensity of the participants responses to the demands were in turn influenced by the support categories that comprise the spokes of the wheels. When all of the support categories were present, the women reported strong resources on which they relied to assist with their health and children. When some of these categories were absent, the participants reported compromising their care in an effort to meet the demands of the situation.

The defining characteristics that are listed under the time line represented participant life events. These life events were not classified as categories because of how they were used and described by the participants during the research. They reflected the types of exemplars that participants shared to describe or clarify questions during the participant interviews. There was not a consistent response pattern in these exemplars and the investigator made the decision to group them under the time line as demands.

The category, level of energy, was designated as a category because of its constant presence throughout the study. Participant descriptions of this phenomenon were reflective of both a support and a demand. Because of its constant presence, they continually adjusted their lives to accommodate the energy fluctuations. Based on the participants descriptions, it has been placed under the time line along with CF and CF exacerbations, growth and development, and transitions as well as in the supporting infrastructure of the wheel. The relationships of level of energy and CF is unclear and requires further study.

This model is preliminary and based on the reported experiences of twelve participants. The categories that were developed were abstracted from descriptions provided by study participants. The classifications applied to the categories were those of the investigator based on participant descriptions. Future research is needed to further explore the phenomenon of interest and other potential categories that might emerge. Additional data are needed to continue clarifying and modifying the relationships of the key categories that emerged in this initial study.

Story Sequences

What follows are story sequences that serve to describe and support the grounding of the preliminary theoretical model in the data. The five story sequences are written in first person and depict the different aspects of the phenomenon under investigation. Each sequence contains the blended experiences of more than one participant as opposed to the voice of one participant. The story sequences serve to further clarify how the model evolved from analysis and why the theoretical reformulation of data shifted away from transition theory. The story sequences are followed by further discussion of the questions that were answered.

Discussion for story sequence one.

Story sequence one is an example of the most common scenario related by the participants. This first story sequence provides several examples of demands on the participant. The demands included family, participant, and disease related experiences. The family demands were the impact that a child's growth and development has on the family

structure and changes that evolved through the family's growth and development. The specific change that occurred was the child becoming more independent and requiring less physical care. This eased some of the constant physical work that makes child care exhausting to mothers. The family's growth was a gradual evolution towards preserving and maintaining the participant's health and quality of life and represents level of energy as a demand.

One of the participant's demands was the transition from work to disability. This major life transition served to temporarily shift the focus back to the long term impact of CF on the participant and her family. What was also important was the experience the participant shared of the process she underwent to develop the infrastructure of the components (categories) of the wheel model to promote health maintaining behaviors. Metaphorically speaking, the components of the wheel that are intact may serve to buffer the shock that comes from demands. When there are major categories missing, the participant has to draw more heavily on existing supports. What may also occur are the development of new supports. Both of these situations are demonstrated in other story sequences.

The support categories illustrated in this story sequence assisted in promoting the health maintaining behaviors. This participant and her family saw themselves as partners in her care. The following exemplar demonstrates the categories of knowledge, personal support, health support, family support, level of energy, home support, and financial. The category of social support was alluded to but was less clearly defined.

Story sequence one.

If I had to do it all again, knowing what I now know, I would not change anything. Life is much richer and more meaningful since the birth of my daughter. I never thought I would ever be able to have a child. I knew CF women were not infertile, but no one ever told me I could get pregnant. No one ever talked to me about birth control. I just figured my cervical mucus was too thick. After I found out I was pregnant, my CF doctor told me he thought I should abort the baby. I was very upset. No one had any recommendations for OB doctors for me. I called around my area and found two other women with CF and they both went to Dr. X who had worked with women with health problems and had been pregnant. He was wonderful. He was very frustrated with my CF doctor because "they" never gave him my lung studies or recommendations for my medicines and how they might affect my baby.

The other way I got information about my pregnancy and CF was over the Internet. My CF doctor could not or would not give me information but there was a lot on the computer. Of course you have to sort through a lot of it, it can be very overwhelming. My CF doctor told me he did not want me doing that. I think it bothered him that I found out more than he seemed to know. Other women who had been pregnant gave me good information on doctors and what I needed after the baby was born–for help and stuff like that. The computer has been a real good source of CF information for me.

Anyway, I have since changed CF doctors. I have to drive farther for my care, but at least everyone seems to try to work with me. They give me more information when I ask questions and listen to me.

Well anyway, I had no problems during my pregnancy. I had some trouble gaining weight but that was all. I saw my (OB) doctor almost weekly during my last trimester. He really watched me closely. I had no problems and no CF related hospitalizations while I was pregnant.

After the baby was born, I wanted to show everyone that I was like every other mother. That I was "normal". While I was pregnant, I felt normal like other women without CF. I had achieved something I was always told was not possible. No one ever talked to me about being a mother. Now I had this baby and I was like others. It was really hard because I was so tired all the time. I found it hard to care for the baby and keep up with my therapies.

My mother came to stay with us for almost a month to help out. I don't know how I would have handled things without her. My parents have always encouraged me to do what I felt I wanted to do. They never discouraged me because of my CF. My mom was able to help me keep up with my therapies and learn to care for the baby. I really wanted to breast feed but I had to give it up. It was easier for my mom and husband to care for the baby and feed her when I wasn't feeling well.

My husband is really supportive. He steps in whenever I need to rest. Unfortunately his family is less understanding.

I try to stay away from children and adults with colds. More than ever, I want to preserve what lung function I have. All our friends know I have CF. But we will not do things with people when they are sick. My daughter is old enough now to know not to bring friends home if they are sick.

I need to maintain my pulmonary function as long as I can and I do not need unnecessary exposure to respiratory viruses. You would not believe how many people, who know my condition, still send their sick children to my house.

Anyway, my in-laws smoke and never seem to respect my need to stay healthy. It makes it so difficult and awkward to have to ask them or the other family members to stay away. They just don't seem to care. When I'm sick or in the hospital, I cannot rely on them to help us out.

I was able to work until my daughter was born. I was very lucky. My boss was very supportive of my CF. When I was sick, he would let me work at home. They would like for me to come back to work. I help them out from time to time. That really helps us out. I'm on disability now because of my CF. It helps to cover most of my CF care. We couldn't swing it otherwise. Even now we don't do anything fancy with vacations or anything. Also, it's hard to plan when I don't know how I'll feel.

My daughter is at an age when she can understand things. I would love for her to be more involved with school and things but I can't commit to taking her places or doing things. She has never complained and tells me how important my health is. Nevertheless,

sometimes I feel badly. What I do is to try to attend things or send things into school that I can do at home.

As much as I enjoy her, summer is hard on me. When she is in school, I can do all my therapies and rest as needed so I can be fresh for her when she gets home from school. I want to be around for all the important events in her life so I really am trying to take care of myself. Sometimes she helps me with my treatments. When I first got my therapy vest, she liked to sit in my lap and feel it vibrate.

I have been lucky. I have only needed to be hospitalized once or twice in the past five years. For a while, I tried to do my "tune-ups" at home, particularly when she was little. But I could not really rest and it was impossible to get all the treatments in by myself. That's the hardest part. You feel bad and need IV's and extra therapies and still have to cook, to clean, and look after a child. It just didn't work for me. I found I got better faster in the hospital. It was hard but my family did better. They really learned how to take care of themselves which helped when I was home. My mother came to help out when my daughter was little. Now she comes less frequently.

My parents are really remarkable. Two of my brothers have CF and a first cousin died of CF. My brothers are older but have been very healthy. Neither of them have children, nor does my brother who does not have CF.

After my cousin died, my aunt and uncle seemed to resent my brothers and me for living. It was very hard on my parents.

My husband and I decided to move across country to another area to try to make a life for ourselves. I guess at the time we felt frustrated by his family's indifference and my aunt and uncle's anger, aimed particularly at me. My cousin (the one who died) and I were the same age only she was much sicker with CF. Anyway, we are glad with the change. We have good friends and support. The only thing I miss about not being back east is my parents.

Both my husband and parents focus on the living. We have never really discussed dying. It's not that any of us are unaware of that, it's just that we are not at that point. Anyone can die at any point but you never really sit down to discuss death. When it happens, it happens. That is how it is with us. We are aware of the difference my disease can make but we deal with more day to day things. While I worry about the future, I'm more aware of the present.

Discussion for story sequence two.

Story sequence two demonstrates the development of the components of the infrastructure of the model as an outcome of a response to a crisis. This exemplar is illustrative of a transition that occurred as a result of a life threatening pulmonary exacerbation. During this period of time, the participant also delivered her baby and was legally transitioning from adolescence to young adulthood. For the first time in her life she experienced the need for periods of home oxygen and intensive home therapy. As a consequence of these very significant changes in her life, this participant quickly identified

the gaps in her life. The categories of demands and to a lesser extent level of energy are represented. The participant briefly references level of energy as a demand.

The participant had to develop home, family, health, and financial supports and these are the support categories represented. The participant quickly learned that being on her own meant having to routinely do her CF care or risk becoming ill. At the time of the interview, her family was assuming major responsibility for the new baby to allow her to recover.

Story sequence two.

We both knew we wanted a baby. I was a little concerned about my CF and how being pregnant would affect it. I "talked" with a number of women on CF chat rooms. They were very helpful. I have "talked" with several of the same women for the past two years. Some you never hear from again and you wonder what happened.

My CF doctor gave me information but he didn't really want me to get pregnant. He felt there wasn't enough known to take the risk.

My mom and sisters help me a lot; they always have. They live next door to us. Someone is always there helping me out. I know I'm very lucky.

When I was pregnant, I had a very bad CF hospitalization. I almost had to be intubated. I was in a long time. They (the CF team) didn't want me to go home but I did anyway (go home). I could do the same care at home.

The first thing I tell other women with CF who are thinking about getting pregnant is to make sure they have a good support system in place ahead of head. You cannot do it

all yourself. Believe me, I tried and it was really bad. You have to learn to accept help if you want to be a mother. Everyone wants to help you out at first; you need to learn how to use them. That's what I tell other women when they ask. You have to take care of yourself if you want to be around for your child. You are no good to anyone if you're too sick to get around and you're going to be sick if you don't do your treatments.

Discussion for story sequence three.

The third story sequence demonstrates the model case. It is an exemplar of a family that was proactive in the participant's CF. The family partnered to preserve the health of the participant. This was an excellent example of the constant presence of CF without seeming to fully dominate the family's life. By maintaining an active stance in CF health promoting behaviors, the family was able to engage in activities that they felt were important. The participant clearly articulated her control over events as opposed to the CF controlling them. This exemplar serves to illustrate the importance and difference in having all the support categories. It also includes level of energy as a support.

Story sequence three.

CF doesn't run my world but it is always there. Besides juggling the care associated with CF, and caring for my children, are concerns about the costs of maintaining my health. My husband and I carry two health insurance policies to accomplish just that. We have two policies. Mine covers the CF pharmacy and CF care. His (spouse) is an HMO that only permits me to see one of their pulmonologists and does not cover a lot of CF care. That pulmonologist (spouse's HMO) has had no CF training

nor does he have expertise with CF. For example, we (the HMO pulmonologist and the participant) had issues with the pulmozyme I was on. He refused to prescribe it for me. I felt it worked, he said it had questionable benefits.

I am being cared for by a pediatric pulmonologist because of his experience with CF. However, when I became pregnant he had limited knowledge and resources to offer me. I found most of my information over the Internet. The CF chat rooms also provided some very practical information.

The importance of adequate health insurance cannot be over emphasized. For women with a chronic illness, it is the means by which to stay healthy. Yes, we have a pre-existing condition and yes we're going to die. But that doesn't mean we're willing to **NOT** remain healthy and productive. We need help to remain healthy, not just treat us when we're ill. The reality is that women with CF are living longer and this has created multiple dilemmas about adequacy of health care coverage.

My husband is my strongest support system. He steps in to help out whenever I am tied up with work or sick. We work together in raising our children together. There is never any question if I need to take time for myself; he always steps in. He has also solicited support through his work place in the form of fund raising. Every year volunteers from his work help out with the various activities organized by the CF Foundation. I believe that the awareness created within his firm has helped him whenever he has needed to take additional time from work to help me.

My parents are my other strong source of support. My sister died of CF when I was in high school. They have always encouraged us to achieve. They never kept us from doing the things we wanted to do because we had CF. They always encouraged us to consider a future for ourselves. My sister continued in college right up to her death. That was her wish.

I have been very healthy and rarely need CPT. I usually only need inhalers. However, with my first pregnancy, I developed several minor complications that required me to be on bed rest. My husband has intermittently needed to take extra time off to care for our children when my CF has been problematic.

I have had no problems continuing to work. Aside from the occasional sinus infection, I have rarely missed work. There are two of us in the firm where I work who have a chronic illness. The nature of my work enables me to work in the office or at home. I know that is not the case with all women. I can pretty much establish my own hours and am fairly independent.

We are fortunate in being able to afford a housekeeper, child care, and lawn service. We also make extensive use of laundry and dry cleaning services. All of these services have enabled us to have more family time and to preserve my health and energy. They are physical energy savers for me. Services like these are worth the extra money if you can manage the expense.

Discussion for story sequence four.

This story sequence serves as an example of the changes that occur over time in the components of the model. For this participant, the life experiences have included, divorce, job uncertainty because of CF, spiritual transition, adult stages of growth and development, and changes with CF symptoms associated with aging. This exemplar demonstrates the varying degree of intensity of demands that can be experienced in response to the demands. It also illustrates the difference in participant response when the components of the model are less well defined and intact. For example, social support became prominent when family support weakened. This story sequence has minimal evidence of home and health support.

Story sequence four.

Cf is a part of me, but it doesn't define me. I am more vigilant with my CF symptoms since the birth of my son. We focus more on day to day activities. We try to plan more family time. For me, I try to decrease exposure to respiratory viruses.

As I've gotten older, the change in my energy level has had as great an impact on my ability to interact socially as any of the other symptoms of my CF. What has helped me are naps and self-pacing with our activities. It is hard to describe how much of a toll these changes in level of energy take. There are times when all I want to do is sleep. Yet, I do not feel refreshed when I wake up. It is much worse when I need a tune-up (pulmonary exacerbation).

What has helped me deal with CF has been my faith. And not just when I am ill. When I was younger, I rebelled a lot. I partied and probably drank too much. I have battled depression my whole adult life. I have been on antidepressants my whole life it seems. I have been on and off antidepressants for the better part of the past ten years.

Anyway, my faith has really helped during my questioning periods, when I have been angry and wonder why me. It has also helped during difficult times when I have been really sick or when my husband decided the CF was more than he bargained for. That was the hardest time. He just up and walked out, leaving me sick and with a young baby. It wasn't like he was unaware when we married. He just said he didn't want to deal with it anymore. My church has been my support ever since. The people there and at work have helped out anytime I need help or a break. My faith has brought me inner peace that I had never experienced before. It is a sanctuary for me. I rarely miss any services or programs. My son is in their preschool program. The community of faith and prayer there has sustained my son and me through some difficult times.

One of my biggest concerns is how much longer I will be able to work. Now, if I feel too bad, I can leave work early. But I am concerned about how I'll manage down the road. I carry health insurance for both of us and it pretty much covers all my CF needs. But I need to work to meet all our needs. My ex-husband provides some help but he is not always reliable. He has taken our son if I need respite help, but I don't know how much I can depend on him. My family lives out of the state and both my parents work. Besides when I lived home, I never had to do much for my CF. I wasn't on CPT, just an

occasional nebulizer or MDI treatment, so my mother has never had to really take care of me.

Discussion for story sequence five.

The final story sequence is an example of the antithesis of the model. This is a case where few components of the model were in place initially for this participant. As a consequence of a series of negative life experiences, the participant describes how she regrouped and learned how to develop a more supportive network. She and her present husband have developed a partnership which reversed the potential outcome. Story sequence five is also an example of the overall toll financial expenses associated with the care of CF can take in discouraging health maintaining behaviors.

Story sequence five.

I did not realize I could get pregnant so when I found out I was, I was really shocked. My CF doctors really pressured me to abort the baby. They did not know what would happen if I maintained the pregnancy. In fact one of the doctors told me I was being selfish in keeping the baby. When I said I would not abort, they then wanted me to have genetic tests done to see if the baby was affected. My whole thing was like is having CF such a bad thing? What is the purpose of the genetic testing–elimination basically if the baby is defective. I mean I have CF and I don't think I am so bad. Whose interest is being served in making the decision about having a baby and having CF? Like no one else has to ask their doctor's permission to get pregnant. I'm a wife and mother who just happens to have CF. Anyway, my doctors did not discuss very much with me. I felt it was more their

opinion. I was able to get more information over the Internet from other moms who have been through the same experience.

Anyways, I had a lot of problems during and after my pregnancy. My husband and I were not getting along and I don't know if that had anything to do with it. After the baby was born, I had to be hospitalized for a long time. My husband walked out. My mom moved in to help me out. She stayed with us for several months. I was in and out of the hospital a lot. Since that first year, I have only been in one or two times.

I met my second husband during that awful first year. He is nothing like the first one. I mean he really takes care of us. But I really didn't want to go through what I went through the first time. So he has been really sweet and patient.

I don't know now, in looking back, How I would have managed without him or my mother. I continued to work, often doing my IV's on my job. I worked different shifts, so I was always tired. I just wore myself out trying to work, care for a child, and keep up with my therapies. I was not much of a wife either. I finally had to go on disability. I really want to work and I miss nursing, but we couldn't afford to pay for my health care. What I miss the most is just staying on top of things. Work stimulates your brain and gives you social contacts. It made me feel worth while and helped with boredom.

Don't get me wrong, I am glad to be able to care for my daughter and step children. But it is nice to get out and have other adult demands and conversations. It was also nice to have money for fun things. We have to be very careful. Sometimes when my daughter really wants something, I have to tell her we'll have to wait on it. She

understands. She helps me with my treatments and stuff. She is very responsible. She fusses at me if I don't take care of myself. She is worth all that I went through at the time. I don't think those who were pushing me to abort understand any of this. I mean, whose interest is being served in making the decision about having a baby?

When others (women with CF) ask me about whether or not to have a baby, I tell them yes–but yes you need to talk with your doctors. You need to talk to others who have done it. You also need to talk it out with your spouse. Mine (the baby) was an accident because I did not know. My doctors did not keep me informed at all. These decisions need to be done with involvement by everyone. There is no way to do it by yourself. Problems always come up. Many of mine would have been reduced if we had all worked together. I don't know if I hurt myself or not but I would do it again, if given the chance, only differently. Women are always going to want to have children and the CF doctors have to learn to work with them as partners.

Summary.

The five story sequences provided excerpts from the data presented in the form of exemplars. The exemplars presented included a model case as well as an antithesis of a case of the phenomenon of interest. The implications for the inclusion of the components of the wheel are the importance of the need to maintain and preserve the women's health. Regardless of the severity of illness, exacerbations are an inevitable part of the disease trajectory of CF and with each episode are physiological consequences of chronic inflammation and infection that over time are irreversible. As a consequence of these

disease events, supports are critical to enable the women to appropriately care for themselves and maintain health promoting behaviors.

The Questions That Were Answered

There were three research questions introduced at the onset of the study. The final section of chapter five specifically examines the three questions that were answered. Chapter five began with a discussion of descriptive data of all the participants. This was followed by story sequences of exemplars from select participants that served to assist in describing the preliminary theoretical model that emerged from data analysis. The last section of chapter five interfaces the descriptive data, the categories from the model with information from all participants, as well as the story sequences to specifically answer the three research questions.

Research Question One: How did women with CF view the impact that pregnancy and caring for children had on their health?

The perception of the impact of pregnancy and caring for children was answered based on information provided from participant interviews, DOII, and NIH-CSCF. The participants framed their discussion of the impact of having a child around two areas: level of energy and disease symptoms. In the theoretical model, Necessary Elements to be a Mother with CF, both of these areas were determined by the investigator to be that of demands as opposed to supports based on the descriptions provided by the participants. The first area, level of energy, was depicted as a category in the theoretical model (Figure 3). The second area was disease focused and included descriptions of disease symptoms,

infection, and treatments. This area was included with life events under the time line on the model.

Level of energy.

Mothers who do not have a chronic illness, experience a change in level of energy when caring for young children. The women who participated in this study all commented about the change in level of energy they experienced after the birth of their children. The range of expression described both in interviews and on the DOII was broad, from present but not interfering with activities of daily living to social isolation and depression.

What made this change in energy level different for the participants in this study was their description of the experience. All participants remarked about the adjustments they made in their lives to deal with this change. The difference was more than needing extra CF therapy because of an exacerbation. For some of the women, even when they were in good health, there was always what was described by the participants as a lack of energy or extreme fatigue.

Several participants experienced what they described as extreme fatigue: "...no matter how many hours I sleep, I never feel refreshed. I am tired all the time. The difference is even more when I need to be "cleaned out" (pulmonary exacerbation). Then I cannot even do simple chores around the house." Manual examination of the NIH-CSCF for this group showed correspondingly lower scores for psychological health, higher expressions of intensity of demand for social isolation, as well as comments during interviews about feelings of depression, loneliness, and isolation.

The link of social isolation and depression to fatigue was not possible in this study because of the small numbers. However, it was interesting to note that the participants with lower NIH-CSCF scores were either on or had been on antidepressant therapy. Secondly, whether the change in energy levels was problematic prior to pregnancy was not explored in this study.

Disease symptoms, infection and treatment.

Nine of the twelve participants reported they had intentionally limited the size of their family based on concern about the potential impact on their CF. All participants reported a change or an increase in CF symptoms and treatments during or following the birth of their children. The discussion of this information during participant interviews was based on retrospective reports as none of the participants was pregnant at the time of the study. The participants made the observation that there was a change or an acceleration of symptoms associated in part with an increase in the number of infections experienced from being around young children. The change in symptoms for four of the participants led to their ultimately being diagnosed with CF.

Two participants stated they experienced a definite decline in health as a result of having children. In both cases the women described more frequent hospitalizations and declining PFT's with each subsequent child. Their NIH-CSCF pulmonary scale subscores had corresponding changes on chest x-rays and CF pulmonary symptoms. The participants reported several contributing factors. These factors were the participants' perceptions of their ability to find time and assistance in caring for themselves.

Two other participants experienced a significant decline in health associated with severe pulmonary exacerbations. Both had contracted multi-resistant organisms which contributed to their decline in health. They did not feel these situations were related to their pregnancy or being a parent. The changes in pulmonary symptoms were also supported by data on the NIH-CSCF.

Linked to the report of CF symptoms was information provided during participant interviews about childbearing and CF. All participants expressed concern about the impact of pregnancy and child rearing on their health and were desirous of making informed decisions based on knowledge shared by their CF health teams. The women in this study found that overall much of their information about pregnancy and CF came from Internet CF chat rooms and/or Internet searches. Additionally, most participants reported limited support from CF health teams. Two participants reported being counseled to abort the baby even though they had "almost normal PFT's". The participants who were followed by high risk obstetricians stated the doctors were open about their lack of experience with pregnant women with CF. However, collaboration between high risk obstetrical and CF teams was reported by the women as being limited and contributed to problems of communication and confusion about CF therapy.

Summary.

Participants reported changes in CF symptoms during pregnancy and in caring for their children. Overall, most participants felt they had done well and had not experienced a decline in health. A decline in health was described by participants who had multiple

pregnancies and children as well as through complications of CF unrelated to being a mother. Again, these descriptions were based on retrospective reports. The study did not attempt to examine or compare the relationship of "normal" CF disease acceleration and aging in women who chose to remain childless.

Research Question Two: What demands were placed on women in caring for their children and their CF?

The demands reported by the participants varied greatly depending on many factors. One participant remarked during her interview that "CF doesn't run my world...it's always there". The demands that were described are found in Figure 3 in the theoretical model. They are listed under the one-directional arrow that represents the time line and are depicted as demand arrows external to the support infrastructure of the wheel. The demands listed under the time line are CF and CF exacerbations, level of energy, growth and development (for both adults and children), and transitions. The level of energy has been discussed under the first question. The solutions to demands described under question two were reported as the supports needed in order to maintain participants' health and care for their families and are discussed under question three.

CF and CF exacerbations.

CF was reported as being constantly present for all participants involved in this research, regardless of illness symptoms. The role it assumed in the participants' lives depended on disease symptoms. The range of reported CF disease symptoms was broad, from chronic sinus disease to double lung transplant, and discussed in detail under

participant demographic information. CF was not viewed by any of the women as dominating their lives. More specifically, while CF was always present "it does not define me". As a way of depicting this concept visually in Figure 3, CF was the inner most shape. However, particularly when CF symptoms were more pronounced because of an exacerbation, CF became a disease demand and was reported to restructure the participants' lives.

There were few aspects of the participants' lives untouched by disease concerns or demands. Issues of sex, sexuality, and reproduction were discussed by 75% of the participants. Sexual and reproductive problems included fatigue, thick cervical mucus, infertility, and irregular menses. Seventy percent of the participants were aware they could get pregnant, however, 55% of the participants did not plan their pregnancies. Two women underwent in vitro fertilization to conceive their children because of problems with thick cervical mucus (Table 2).

Another important concern of the participants about disease demands centered around the financing of health care. CF therapy involved expensive medications and treatments necessary to maintain the participants health. Costs involved in CF health maintenance and exacerbation were described by the participants as immense. Additionally, costs tended to proportionately increase with participant age because of acceleration of CF related symptoms. As a consequence of these costs, there were three financial concerns expressed by participants related to CF and CF exacerbations: employment, health insurance, and costs related to health care. Information detailed under

the participants' demographic data and the story sequences discussed earlier in this chapter alluded to these concerns. Because of the consequences of inadequate health care coverage and the importance of the inter-relationships of disease care, health maintaining behaviors, and health care costs, the following discussion provides a more detailed account of the problems reported by participants.

The discussion will begin with the concerns expressed about employment. The cyclic nature of CF exacerbations provided challenges to employment. All participants expressed an interest in working, however, CF therapy, fatigue, and CF symptoms prevented 66% from achieving this objective. The primary reason participants stated they found it difficult to remain employed was frequent absenteeism. When CF therapy required more than oral antibiotics participants said they felt too fatigued and ill to work. Sick time away from work entailed one to three weeks of increased treatments. "When I'm healthy I can spend six to eight hours per day doing treatments. When I'm sick, I find I spend all my awake time caring for myself. Even when I'm healthy I find I need an afternoon nap to be able to take care of my children when they return from school". In reality, few work places could support CPT, aerosol treatments, intravenous antibiotics, or tube feedings. Additionally, employment benefit packages were not designed to handle extended periods of time off from work for home or hospital therapy during exacerbations.

Participants who were employed reported they were fairly healthy and experienced few CF symptoms or exacerbations. Less than 2% of these participants reported they had required hospitalizations for problems. Participants employed more than 20 hours per

week reported relying on work flexibility, family, and community support to enable them to continue working.

The second area reported by the participants were issues dealing with health insurance. Health insurance issues primarily centered around financial coverage for medications and therapies. Insufficient health care coverage seriously impacted participants' ability to engage in health protecting behaviors such as CPT. CPT was reported by participants difficult to administer without a second person or a mechanical device called a therapy vest. Only one participant reported having insurance that paid for home based manually (hand) administered CPT. The therapy vest offered autonomy, however, only a small number of insurance companies were willing to purchase the device because of an estimated cost between $10,000-15,000. Participants saw themselves as having few options to administering CPT when help was limited.

Participants who required CPT more than two times per day reported missing therapies when home, family, and health support was insufficient or unavailable. Less expensive CPT options existed (e.g.–flutter valve), however, these options were viewed by some participants as less effective than manual therapy. Importantly, several participants stated that while less expensive devices were offered by CF teams, the approach in terms of education and training with the devices was viewed as inadequate to provide a level of comfort for use at home. As a consequence of the lack of sufficient health support and follow up, the CPT devices were unused or CPT was missed. What

participants reported as enabling them to maintain and sustain their home therapies were health care teams who partnered and followed up to explore viable home options.

Health maintaining therapies and medications that were not fully covered by health insurance (or a lack of insurance) were reported by several participants as contributing to a decline in health. The problems these participants reported were high insurance co-pays or deductibles. There were also problems with health insurance companies approving the substitution of less expensive drugs which were reported by the participants as contributing to increased CF symptoms. This was most problematic for participants on pancreatic enzymes with resumption of symptoms of malabsorption,

Participants who had to pay high health insurance co-pays for CF care, found it difficult to purchase medications and therapies. Many reported waiting to seek treatment for symptoms because they could not afford costly therapies. This group also reported the highest intensity of demands on the DOII on family finances and alterations in family lifestyle. The study did not attempt to examine formal relationships between the variables of health insurance, CF care, and hospitalizations.

There were also issues with health insurance policies related to specific health care coverage. Problems with having to make appointments with non-CF trained pulmonologists designated by health insurance companies was another concern expressed by the participants. There were a number of health insurance policies reported by the participants that designated which specialists would be paid for, the type and amount of

medications covered, what tests would be paid for during office visits, and the choice of hospital versus non hospital based care.

The final area discussed by the participants about CF and CF exacerbations were costs related to the direct provision of health care. Costs of medications and therapies for daily CF care were reported as often exceeding $10,000 per year. Additional costs were added during intensive therapy for CF exacerbations whether the participant was hospitalized or received out of hospital based care (e.g.-home based).

The advantage and disadvantage of hospital versus out of hospital therapy for pulmonary exacerbations was discussed by the participants. They commented on the insurance industry's trend towards promoting non hospital based therapy for CF exacerbations, in an effort to contain health care costs. Three participants who had experienced home therapy, expressed the difficulty of caring for young children and home intravenous (IV) therapy, increased CPT, and nutritional supplements. "When I am sick, my CF care can exceed eight hours per day. When I'm in the hospital, I worry about what happens at home but I can rest and concentrate on getting better".

There were participants who preferred having the option of non hospital based therapy and preferred having the option of being treated at home. "The hospital is two hours away. At least I can stay on top of problems that come up. My family doesn't have to arrange for sitters for children to come see me". What was viewed as critical was to have a CF health team and health insurance company who worked with participants to explore the best health care treatment options.

A second but less traditional area where participants incorporated non hospital based therapy was in the work place. Non-hospital based therapy permitted IV therapy to be done at the work site in an effort to decrease absenteeism. Two participants had administered IV medications at work and felt it helped them in remaining employed. One of these participants commented however, that when she did CF therapy at her place of employment, she had to work additional time to make up the hours lost in delivering CF care. Realistically, participants felt that few work settings were conducive to support CPT or supplemental nutrition in the provision of space, privacy, and time to adequately deliver treatments.

Demands of transitions and growth and development.

Incorporated under the time line in Figure 3 of the model Necessary Elements to be a Mother with CF, were two other demands: transitions and growth and development. Participants constantly struggled with a number of factors related to these life events including changing interpersonal relationships, aging parents, and reliable child care help. Another important disease demand discussed by the participants was learning to deal with the long term consequences of having a chronic illness. The implications of the effect of problems in these areas was profound in the context of advancing CF disease. The story sequences discussed earlier in chapter five provided concrete evidence of what can happen when changes occur. Changes in life events had the potential of eroding or providing the support base so necessary for maintaining health and caring for children.

Like other chronic illnesses, CF exacerbations were cyclical; unlike individuals with physical disabilities, the need for help with self care and in the home was not always predictable nor constant. "It's not like I need to ask for help once or twice. There are limits to how much help you can get and how much you can ask others to do".

The story sequences provided examples and discussed changes in interpersonal relationships with families and partners. Most of the changes in relationships came about as a result of the realization that despite the desire to be independent, CF care to maintain health was an important priority. The participants in this study discussed the process of discovery that led to this point and the role their parents and relatives maintained or re-established in their adult lives. Participants reported their families to be equally involved and at times more involved than the partner. The importance of family involvement was expressed as crucial in enabling participants to meet disease demands and changes in life events in caring for themselves and their children and is discussed in more detail in the next section.

Changes in life events also extended into the area of socialization for both the participants and their families. All participants reported some degree of change in social engagements. These changes were primarily based on how they were feeling. No one reported feeling socially ostracized because of their disease. Participants did report being self-protective by actively discouraging contact with potentially sick adults and children. "I need to maintain my pulmonary function as long as I can and I do not need unnecessary exposure to respiratory viruses. You would not believe how many people, who know my

condition, still send their sick children to my house. My son seems to understand how it is... he never complains when I do not feel well enough to attend his activities at school". Another participant noted "there are so few days when my sinus disease doesn't cause me to be in pain. When I am feeling good I don't want someone else causing me to get infected again so quickly. I am not even nice about it anymore".

Five of the participants reported experiencing significant enough CF symptoms, most notably a drop in level of energy, to seriously curtail social activities outside the home. They expressed the need to prioritize activities and conserve energy for time with their children. "I love when school starts, then I have enough time to throughly do my treatments, take naps, and enjoy my children when they come home from school". An important developmental milestone of a child beginning school then became a support and eased the demands of self care with the provision of time to do health maintaining CF therapies.

Summary.

The demands of needing socialization for emotional and developmental reasons as well as to sustain personal support, coupled with the need to be protective of their personal health, created unique dilemmas for the participants. The demands discussed in this section are illustrated in the Figure 3 model, Necessary Elements to be a Mother with CF, as external to the wheel and are described under the time line. The demands perceived by the participants that are presented in this section show how the women struggled with how best to preserve their health. Participants had to juggle children's school and social

activities with their own personal level of wellness. The intensity of demands associated with caring for CF created significant stressors that impacted family dynamics and interpersonal relationships. Added to the myriad of family issues were the demands associated with the costs of financing participants' health care. The socio-cultural and medical chasm created as a result of advanced medical technology was reflected in the inter-relationships of disease cares, health maintaining behaviors, and health care costs.

Research Question Three: What supports did women with CF perceive as needed to care for their children and manage their disease?

When transitional changes occurred for the participants (i.e.- changes such as moving from employment to disability, moving away from home, or divorce), supports had to be reassessed. While there were many concerns expressed about the demands of having CF and being a mother, participants were also able to share ways they dealt with these challenges. The last research question discusses the solutions proposed by the participants in attempting to meet family and illness demands.

The final research question explored with participants what supports they perceived as needed to meet the demands of caring for their children and in continuing to care for themselves. All the participants discussed the ongoing process of learning to prioritize–"how best to be involved and when to let go. After the birth of my daughter, I had to balance caring for myself and learning to care for another".

Even with prioritizing events to meet family and personal needs, supports assisted participants in achieving what was described as balance in their lives. All participants

addressed the need to learn how to "ask for help" in order to balance family and disease demands. "The first thing I tell other women with CF who are thinking about getting pregnant is to make sure they have a good support system in place ahead of time. You cannot do it all yourself. Believe me, I tried and it was really bad. You have to learn to accept help if you want to be a mother."

In addition to addressing the need to learn to ask for help to balance family and disease demands, the participants identified supports they recommended as useful for other mothers with CF. The supports participants perceived as needed to care for their children and manage their disease emerged to form the six categories described in the preliminary theoretical model, Necessary Elements to be a Mother with CF, found in Figure 3. The story sequences presented several concrete examples of supports that illustrated the categories.

The remaining discussion provides a more detailed look at the support categories. Four of the categories involved personal support to the participants: family, social, health care team, and home support. Two other support categories, financial and knowledge, influenced the kinds of health care services the participants received. The final category of support, level of energy, has already been discussed but influenced what participants could accomplish and their perception of well being.

Personal supports: family, social, health care team, and home.

Personal supports were described as the people, situations, and things that enabled participants to meet the demands of family and disease. One participant put it succinctly

when she stated that "I found through experience the limits to the kind of help one can reasonably expect and the boundaries to what others will do". Four categories of personal support were identified by the participants as important: family, social, health care team, and home.

Family support, of all support systems described by the participants, seemed to most directly influence the women's perceptions of success with parenting skills. What participants viewed as critical to their children's overall well being was minimum disruption in the lives of their children. The participants reported relying heavily on their spouse or a family member to "step in" when they were not feeling well or needed to be hospitalized. "Women with CF who are planning to have children need to have a plan in place with people who can respond with a moments notice. These things need to be lined up before problems emerge so everyone knows what to do." The relevance and importance of this family support system seemed strongly connected to the need to have stable child care.

The development of a family support system coupled with the concern about their children maintaining a "normal" home life, co-existed with the need of the participants to preserve health and well being. "My biggest worry is what if something happens to me. Will anyone else give them the care and understanding they (the children) need? When you are healthy and feeling good, you don't want to think about anything else. But you need to plan now, when healthy, how and who will take over to do things; you need a quick response time. I am a lot more aware now of life and things going on around me. While I

worry about the future, I'm more aware of the present–I'm a lot stronger because of the experience. I'm not afraid to get help when I need it. I now know when I need a break and care for myself. So much depends on this".

The participants and their children were representative of a variety of ages and the process of understanding the need to balance "self" and children emerged in a manner that was reflective of a developmental time line. Initially, participants expressed the difficulty of asking for help until an emergency developed. With the passage of time, they began to see and feel the difference supports made in their over perception of control and well being. The participants began to describe more proactive behaviors in developing and maintaining personal supports.

Personal support systems were reported by the participants to mature over time. There were two reasons for this: 1) children became more independent as they grew older, and 2) child care providers tended to stabilize. The participants were able to identify more stable and reliable individuals for child care. However, there continued to be limitations to personal supports because of changes in the lives of those attempting to help the participants.

What was reported by the participants as being the most helpful were children becoming more independent and requiring less direct care. As children matured, some were able to help with the participants' CF treatments. None of the children in this study were reported as assuming any primary responsibility in their mothers' CF care, regardless

of financial circumstances. The participants continued to use adult supports for CF treatments.

The next category was social support. Social support was described by the participants as providing additional personal support. This form of support for the participants went beyond family support and was important to their children and family in maintaining normal activities (see Table 7). These supports did not always provide immediate or respite relief to the family, as much as helping out in other ways. Particularly in situations where the mother's disease was more advanced, social supports aided in maintaining a more normal pattern of family life for children. "It has always been difficult for me to ask for help. However, as my disease has progressed, I am now more active in doing the things I need to do to stay healthy. Fortunately now my son is old enough to care for himself. My husband has remained very traditional. While he was aware of my disease before we got married, I was very healthy. Up until several years ago, I was able to do everything. Not anymore, I am too tired. When others are able to provide hot meals or even run errands, it makes things less stressful. My husband and son do very little to help out around the house–cooking, cleaning, laundry or anything."

The participants expressed the desire that their children experience as "normal" a life as possible. There were a number of social supports that were reported as helpful by the participants. Social supports that were viewed as helpful ranged from hot family meals when the mother was sick to transportation for school and extracurricular activities.

Another support that was important was for the participants to remain involved with their children's activities but having the expressed freedom to withdraw at any time

when tired or ill. "There are times when just mopping the floor is exhausting. Juggling the children's activities can be overwhelming. Having to let go of things, particularly hardest with just the basic things, but I know when I need to let go. When I'm sick or one of the children is sick–one parent needs to be available. Everyone has to help out. My husband's aunt lives next door. She seems to know when to step in. She keeps an eye on things when I'm sick. I know I can rely on her."

The final area under the category of social support was indirectly related to the participants. This form of support involved community or job related support in fund raising for CF activities. A number of businesses regularly participated in CF fund raisers. Additionally, communities had directly raised money to provide CF care for some of the participants (e.g.-- lung transplant). All of these activities helped to relieve some of the physical and psychological demands encountered with having a chronic illness and being a mother.

The fourth personal support category identified was the health care team. The role of health care teams in providing personal support varied widely. Several concerns have been identified under the section discussing demands. Some of the participants experienced issues around use of CF versus non CF specialists. Problems also arose with communication between the participants and their health care team as well as between health care specialists created opportunities for fragmentation of care.

The basis for the reports of communication problems was examined from responses on the DOII and participant interviews. Participant responses to questions about

support from the health care team were found to be incongruent. Overall, on the DOII, the participants reported confidence in the skill and care received from the CF team, but when interviewed did not perceive them as providing support. The participants who continued being followed by the same CF team reported the strongest support; those who had transitioned from a pediatric to adult CF health care team reported feeling less support.

In part, the differences expressed on the DOII and in participant interviews related to the definition of support. The participants defined support provided by the health care team as more symptom based. "I'm satisfied with how my illness is being managed and am confident it will be correctly managed in the future. However, I've had problems with specialists who only look at symptoms related to their area and did not look at me as a whole. I found my care very fragmented. During my last two pregnancies the CF team provided very little support or help. They made it clear I had done the wrong thing. The education and information they did share about CF and being pregnant seemed to be inconsistent and uneven. I felt confused and abandoned". The participants were looking for and needed support that included their emotional and physical needs, as well as added to their knowledge about CF. In essence they were looking for a partnership based on an honest working relationship with their health care team.

The final category of personal support perceived by the participants as needed to care for their children and themselves was home support. Home supports were defined as those things that enabled participants to "keep" house, care for their family, and care for themselves (Table 7). While daily household routines for healthy women may be

accomplished effortlessly and unnoticed, at times achieving the least strenuous task was difficult for women with a chronic illness.

The importance of support in the home has been touched upon under the section discussing demands. There were three home supports described by the participants: 1) someone to help clean house, laundry/dry cleaning, and do outside yard work, 2) the provision of reliable child care, and 3) assistance to free time for them to care for their health. What these three types of home supports enabled the participants to do was to conserve their energy for caring for their children. They helped to escape the feeling of guilt associated with trying to "do it all". The participants also observed that the flexibility to be able to participate or withdraw from household and child care responsibilities promoted positive mental health.

A home support system that stayed attentive and responsive to the participants' needs aided in maintaining a more healthy outlook. An attentive and responsive support system discouraged women from postponing their health needs and missing therapies. Participants with these components reported they were less likely to "tough it out" in seeking medical care for changes in CF symptoms. "My husband is usually the first to notice my increased cough and fatigue. He immediately steps in and arranges for things to be done around the house and for the children to allow me to rest and increase my therapies". The participants who reported supportive environments also had lower scores on intensity of demands on the DOII for items related to support.

Conversely, the participants who reported a less supportive home environment had higher scores on intensity of demands for items related to this area. The consequences of a lack of access to respite help in the home were compromises to their health in an effort to keep the family functioning. "My husband and son do not feel they can help me in any way. When I am sick, I have no way to increase my CPT, so I usually only increase my medications. Fortunately, I'm rarely sick but I know I wait until I'm really sick before seeing my CF doctor. Home therapy is not an option for me. My CF team and insurance (health) have tried to encourage me to do home therapy. But how can I take time out to be sick with a house and a family to care for". Another participants who reported limited home supports observed that "her dad is no help (the child's father). I cannot rely on him to help even if I ask. Fortunately I don't get sick that much, but I worry what will happen if my health deteriorates. My parents live too far away and friends can't always step in the way families can".

The benefits to home supports were that they enabled participants to take time to care for themselves. Overall, the participants could manage without them, however, they provided the needed time to adequately and appropriately do health maintaining therapies. "My attitude has changed to be more aware of caring for myself and living. While fatigue is not an issue for me, I try to take advantage of outside supports. I can afford to have help in other ways with supports such as someone to clean the house, laundry and dry cleaning services, and day care. These services free up more time to spend doing things I want and need to do."

In summary, personal supports enabled the participants to meet the demands of their children and care for their disease. These four categories of support provided the participants with opportunities to remain active in their children's lives and in keeping family disruptions to a minimum. They enabled the participants to have the opportunity to experience fully the role of being a mother.

Financial support.

The category of financial support moved away from personal supports and dealt with monetary problems related to maintaining CF care. Inadequate health care coverage presented difficult moral choices for participants about obtaining CF care and in creating financial hardships for families. This category was dominated by reports of problems that arose with health insurance.

Health insurance was a form of financial support that enabled participants to obtain the necessary CF centered care for medications and therapies to maintain health. The solutions to these two needs of the participants formed the basis for this category, the first being the importance of having adequate health care coverage to maintain health and the second adequate health care coverage to receive CF based health care.

All participants acknowledged the crucial role of health insurance in the life of people with a chronic illness. Unfortunately, there were no clear answers to this complex problem. One participant recently experienced the cancellation of her group health insurance. Because of her part time employment, she was not eligible for benefits through her employer and has been unsuccessful in obtaining an affordable individual policy. Other

participants continued to work to maintain health insurance in order to prevent this very problem; individual policies were too difficult to obtain or financially out of their means.

For other participants who could afford it, the problem of adequate health coverage was resolved by obtaining more than one health insurance policy to cover health care costs. Some participants carried insurance policies for themselves and were also covered by their spouse's insurance. "We have 2 policies. Mine covers the CF pharmacy and CF care. His (her spouse) is an HMO that only permits me to see one of their pulmonologists and does not cover a lot of CF care. That pulmonologist (covered by the spouse's HMO) has had no CF training nor does he have expertise with CF. For example, we (the HMO pulmonologist and the participant) had issues with the pulmozyme I was on. He refused to prescribe it for me. I felt it worked, he said it had questionable benefits. Later I found out that it was not approved to be covered by the HMO".

In summary, health care insurance was viewed as a financial support, to provide the means necessary to help women stay healthy. The health insurance had to have coverage that specifically addressed the special needs related to CF care. This type of health coverage enabled families to be free from the stress of major unexpected health care costs. "Yes, we have a pre-existing condition and yes we're going to die. But that doesn't mean we're willing to not remain healthy and productive. We need help to remain healthy, not just treat us when we're ill". The reality that women with CF were living longer created multiple dilemmas about adequacy of health care coverage.

Knowledge.

The final category of support dealt with communication and knowledge. The participants reported three important supports in relation to receiving adequate knowledge and maintaining open communication: 1) the provision of information about CF as it pertained to being pregnant and being a mother, 2) keeping in touch with current information about CF research, and 3) serving as a communication link with other mothers with CF.

The problems with communication with CF health care providers was discussed under research question two and in the story sequences. Two participants reported changing CF doctors because of negative experiences in trying to obtain and exchange information about CF care. Others tried to arrange clinic appointments with members of the health care team they felt would provide them with the best information. Participants felt that open communication with their health care teams facilitated obtaining the necessary information that enabled them to receive better care.

The use of personal computers and the Internet broadened the scope of contacts and information for participants. For most participants it was described as an essential source of information about CF and CF care. All but one participant in this study actively used the computer to access the Internet.

Use of the Internet for Cf related purposes by the participants ranged from daily to weekly. While the computer provided many valuable services, all participants were clear as to its limitations. The limitations of computer technology were similar to the benefits it

offered. Information was described as overwhelming and required careful "triaging and assessment". Secondly, while knowledge of the latest advances assisted in discussing options in CF care, the information that obtained was complex.

The participants who routinely used the computer for sources of CF information stressed the importance of partnering with a health professional to discuss the information. The participants saw the partnering as a way to develop a plan of care appropriate for each individual. Unfortunately, some participants stated they encountered negative and at times hostile responses from the CF team when approached about information obtained over the Internet.

Lastly, communication with other individuals with CF over the Internet was reported as carrying all the risks that other disease specific chat room groups have reported. For example, little information was known about individuals who communicated through chat rooms. Conversations frequently turned into macabre and morbid topics. For healthy individuals with CF, such conversations were reported as providing little if any relevance or support.

Summary.

The third research question explored with participants how they were able to meet the demands of caring for children and their own health. The participants expressed the importance of the use of supports for help in balancing family and CF.

Family, social, and health care team supports enabled participants to maintain an active role in their children's lives. Home supports assisted in maintaining home

operational systems and provided time needed to care for their disease. Health care insurance ensured that participants had health care coverage to maintain health as well as for CF exacerbations. Lastly, knowledge and communication permitted the participants to stay abreast of the latest CF information and research as well as a means to keep in touch with other mothers with CF

Chapter Summary

Three research questions were initially explored to understand how women with CF incorporated caring for children with caring for their CF. The questions specifically explored the women's perceptions of the impact of pregnancy and child care on their CF, demands of caring for children and CF, and needed supports. From responses to research instruments and participant interviews, categories of responses were developed. The final level of abstraction of the research data was the development of a theoretical model grounded in the data (Figure 3). The scaffolding developed under transition theory at the outset of the study was examined to see how well it fit the model that ultimately emerged through data analysis.

Chapter six discusses lessons learned from the research. The chapter goes on to outline what the next questions should be for future studies.

Chapter 6

Discussion

This journey began in chapter one with a discussion of the tremendous impact advances in medical technology had on women with chronic illness. The initial aim of the research was to link outcomes of pregnancy and child rearing with shifts in disease severity. However, what ultimately emerged as the research proposal explored how women cared for children and continued to manage their disease. To understand the impact these advances had on the participants, the women responded to both a formal questionnaire and semi-structured interview. What evolved from the research process was further evidence of the impact of the chasm between knowledge, technology, societal expectations, and the lived experience of women with CF. The theoretical model developed from the data reflected social, medical, and economic themes of how these women worked to sustain and maintain family life and a chronic illness entering the twenty first century.

Lessons Learned

The cultural norm in most western societies encourages reproduction and motherhood as a valued female role (Leifer, 1980). The impact of the gap left for the participants between increased life expectancy because of medical technology and the socio-cultural expectation of an early death created multiple dilemmas. The participants viewed themselves as "normal" women wanting and having children. They saw themselves carrying out normal roles, except they "happened to have a chronic illness". On the other

hand, society viewed the participants as "aberrant". The participants made the observation that others saw them as "having outlived their life expectancy and now were selfishly desiring children". Specifically, the medical community was questioning their desires and motives in having children.

Three important lessons were learned from the participants that all related to the chasm created by advanced medical technology and socio-cultural norms. The three lessons were: 1) the disconnect in reproductive cultural norms for healthy women and women with a physical disability or chronic illness; 2) the challenges with the type of support provided by the health care system to mothers with a physical disability or chronic illness; and 3) the disparities between medical, social, and economic expectations of being a mother and having a chronic illness. For each of the lessons, research findings will be compared and contrasted to other studies.

Lesson one.

One of the first lessons learned was the disconnect between the reproductive cultural norm for healthy women and for women with a chronic illness or physical disability. The social response to reproduction in the participants in this study was complex and interwoven with concern for the women's health and long term outcomes. Like other women who fall outside the accepted cultural norm, there was a general lack of recognition and support for the reproductive experience regarding maternal identity and motherhood (Corbin, 1987; Ingram & Hutchinson, 2000; Lipson & Rogers, 2000; Miall, 1989). What is disturbing is that social stigmas continue to exist and to persist for mothers

whose life experiences fall outside culturally accepted norms of reproduction. Reproductive literature over the past decade on women with physical disabilities or chronic illness continues to demonstrate general discouragement and a lack of support by health care providers.

Despite the negative views that some participants in this study experienced from health care providers, the women valued being a mother and were highly motivated in maintaining their health. The high priority motherhood had in their lives was reflected in the theoretical model, Necessary Elements to be a Mother with CF, that emerged through data analysis (Figure 3). This study reflected findings similar to those reported in a study on HIV-positive women of the life-sustaining role children had in their lives (Ingram & Hutchinson, 2000). The participants in both the HIV study and in this study with CF identified their children as the reason for being and the impetus to continue to fight to survive and remain healthy.

The fluctuations in levels of energy challenged participants as they tried to meet family demands. The amount of energy described as being expended to maintain health and health sustaining treatments was immense. Some of the strategies described by participants to meet disease and family demands were also described in a study by Wuest (2000) as repatterning care. Wuest (2000) examined how healthy women, in response to family caregiving demands, used a process of repatterning or reorganizing caring activities as a means by which to reduce or overcome the negative effects of caregiving demands. What she described were processes employed by healthy mothers to develop supportive

family supports, use substitute (child/adult) care effectively, and care for themselves. These same processes described by the mothers in Wuest's study were supported by the findings in this study. What mothers in both studies were attempting to accomplish was a proactive approach to management of their families.

The participants recognized the difficulties of achieving the social ideal of being a good mother and balancing the demands of their CF. With a CF exacerbation, the participants' health came first. On the other hand, when participants felt good they were able to refocus on the needs of their children. This finding was different from other studies where mothers stated they always put the health and needs of their families before their own, even when it potentially jeopardized their health (Corbin, 1987; Hackl et al., 1997; Ingram & Hutchinson, 2000; Lipson & Rogers, 2000; Miall, 1989; Thorne, 1990). The importance of respite care with seamless substitute care for their children was critically important to maintain balance and a normal home environment.

The concept of normalization has been described in the context of parenting a child with a chronic illness (Deatrick, Knafl, & Murphy-Moore, 1999). Attributes associated with normalization are: acknowledges condition and its potential to threaten life style; adopts normalcy lens for defining child and family; engages in parenting behaviors and family routines that are consistent with normalcy lens; and interacts with others based on view of child and family as normal. Aspects of normalization might be useful to the present study. For example, the concept deals with family management and the behaviors

families acquire in an effort to reconstruct their lives around the chronic illness to be as normal as possible.

Certain aspects of the attributes of normalization fit with the behaviors described during participant interviews. However, what was problematic with the concept to current findings was that the mothers in this study were the ones disrupting normal family patterns. At times participants felt unable to mother effectively because of the demands of their disease. The participant's level of energy dictated the likelihood of her ability to engage in management behaviors consistent with normalcy.

Lesson two.

A second lesson learned was in the chasm between the socio-cultural expectations of being a mother and a woman with a chronic illness and the support provided within the U.S. health care system. The similarity of experiences between the research findings in this study compared with the literature, particularly given the age of some of the studies, is striking and concerning (Corbin, 1987; Hackl et al., 1997; Ingram & Hutchinson, 2000; Lipson & Rogers, 2000; Miall, 1989; Thorne, 1990). For example, Thorne (1990) in her study examining the experiences of chronically ill mothers, identified interactions that were related by women with physical disabilities such as multiple sclerosis, between their chronic illness and mothering. Thorne outlined four interrelated mothering concerns based on performance, availability, dependency, and socialization. What she and the participants discussed was the inability of health care and health care services to meet the demands associated with being a mother and having a chronic illness. Unrealistic appraisals by the

health professionals were thought to be a product of specialization and fragmentation in health care services. Now, a decade after the initial publication of the research by Thorne, women are still reporting similar kinds of problems (Lipson & Rogers, 2000).

A number of health care system factors impacted the level and quality of care. Lipson and Rogers (2000) found in their study of mothers with physical disabilities that high-risk obstetricians were primarily trained to work with problem pregnancies. As a result, there was little focus on the mother's physical condition (for the women with physical disabilities) except as it affected the baby. Another problem was in communication between physicians and specialists. There was a general lack of seeking consultation or additional information with the medical staff. Similar problems were reported by participants with CF in this study.

Medical advances with improved technology and life expanding therapies are enabling children entering the twenty first century to mature to adulthood. Medical technology has enabled girls to have opportunities not possible in prior generations of girls with CF. However, what is equally and perhaps more concerning, is that the current generation of young women who have grown up with their illnesses, are reporting the same kinds of problems detailed in studies from a decade ago.

There were some differences between the studies cited and this research. Some of the problems described in studies of mothers with a physical disability such as multiple sclerosis, were not described by the participants in this study (Corbin, 1987; Nehring & Cohen, 1995). These studies described problems with adapting equipment to care for

children and the associated guilt with not being able to do things with their children because of fatigue and impaired mobility. The participant's social construction of being a mother actually incorporated their CF. This may in part be due to having early on internalized CF as part of their self identity. Because CF symptoms have been present since an early age (regardless of age at diagnosis), the role of becoming a mother focused more on issues of how to maintain health sustaining behaviors.

The participants in this study expressed frustration by the limitations imposed by CF but never anger nor feelings of inadequacy because of the disease. What continued to permeate the experience of being a mother with CF were issues of performance because of lack of energy, availability because of the cyclical nature of CF exacerbations, socialization, supports, and health care.

Lesson three.

The third lesson learned was from the categories that emerged through data analysis. The categories that emerged from analysis of participant responses clearly reflected the chasm created by the disparity between social, medical, and economic expectations. This disparity required rethinking social history for individuals with CF. For the participants, the disparity created three primary challenges: issues of child care, long term monetary costs associated with having CF, and maintaining belief in oneself–"balancing what you have with what you can do". These were discussed in detail in chapter five.

The three primary challenges experienced by participants were not so different from mothers who did not have a chronic illness. However, in contrast to "healthy mothers", the participants were extraordinary in forcing a rethinking of social history medically, socially, and economically with regards to: 1) having a chronic illness, 2) extending the boundaries of life expectancy, and 3) being a woman of child bearing age. The demands the participants described through their responses to the questionnaires and interviews were more issues of time management and health maintenance. Health care providers and agencies governing health care coverage were described as being unresponsive to the needs of participants. Many of the solutions that were described by the participants evolved through trial and error because of a perception of lack of adequate supports.

The perceived unresponsiveness of health care providers to the needs of the participants forced them to turn to other avenues for support and information. A unique form of support and information that emerged from the study and was described by the participants as highly effective was the Internet. This too may be another example of the gap between technology and the current cultural climate. Irrespective of the nature or origin of these gaps, 92% of the participants reported the Internet as a critical and positive form of support. Because this study primarily focused on women with computer skills, it was not possible to evaluate resources used by non computer literate women.

In summary, societal values in general are placed on having children; having a child is considered basic to a woman's life, seen by some as a necessary element to completing

and fulfilling one's life. The participants in this study felt they were responding according to societal expectations by viewing reproduction as socially and psychologically desirable. They expressed the belief that by having children, they, like other women, would fulfill the personal and social expectation of being a mother.

What Are the Next Questions?

Participants in this study were self-selected and research findings were based solely on reports from their experiences. The intent of the study was not to generalize to all mothers who had CF. In considering the next phase of the study, there were a number of additional questions that were raised based on participants' responses.

Several other areas were touched on by the participants that were outside the scope and intent of the phenomenon being explored in this research. The additional areas that emerged from participant interviews included: 1) spirituality, 2) the relationship of level of energy, social isolation, severity of illness, and use of antidepressant therapy, 3) use of the Internet and the role it has for individuals with chronic illness, 4) comparison of disease outcomes of women with CF to those who chose a career trajectory, 5) the relationship of the adequacy of financing of chronic illness and the impact it has on health maintaining behaviors, and 6) the role of the concept of time.

The first area was spirituality. A number of participants discussed the importance of spirituality in their lives, as a form of personal support. The role that was described was variable as to importance and magnitude. Because of the small sample size, it was not possible to explore the significance of spirituality nor the relationship it has to the model.

A second area that emerged from analysis of data was level of energy, social isolation, and use of antidepressants. What was observed in the data was that participants who expressed a high degree of fatigue (or a low level of energy) and social isolation were generally sicker. However, they also reported use of antidepressant therapy to control symptoms of depression. No one in this study reported suicidal ideation or gestures. Again, because of the small number of participants, it was not possible to go beyond describing the patterns that were seen.

Another area to be further explored was use of the Internet by individuals with chronic illness. This study attracted predominantly women who were Internet users. A number of uses, specifically for support purposes, were described by participants in this study. What is not known is the significance of the role and use of the Internet that is emerging for people with a chronic illness.

This study examined mothers who had cystic fibrosis. However, a number of women with CF chose not to have children but opt for a career path. Current CF literature was unclear as to differences and/or similarities of disease outcomes between these two groups of women. Further research comparing outcomes of disease for women with CF with children to those who opt not to have children and follow a career trajectory would be important for women in planning their futures. How do women with CF manage a career and their disease and how does this compare to the demands of a family?

A fifth area discussed by the participants for this study was the question of adequacy of the financing of chronic illness and the impact it has on women's ability to

sustain health maintaining behaviors for CF. This was a social policy issue and has a significant impact on the economics of financing health care. The participants discussed several concerns that involved obtaining appropriate medications, treatments, and use of specialists. The problems involved with these issue are very complex and their impact is far reaching. Further research would assist legislators and the insurance industry to make knowledgeable decisions.

The final question is the emergence of the concept of time in the theoretical model, Necessary Elements to be a Mother with CF. Based on the findings from this study, research is needed to continue exploring the challenges of balancing caring for children and CF and to answer the question of where the model of concept of time fits into all of this. The outcome is to situate within the normalcy of wanting to achieve the experiences of motherhood, the participants' difficult social, medical, and economic themes in terms of what it takes for these women to sustain and maintain family life with a chronic illness. The goal is to avoid framing the target phenomenon from a sociologic theory of deviance.

The next challenge is to develop a meaningful and rich theory that represents the many experiences of mothers with CF. The direction of the clinical usefulness of the model needs to remain focused on the inter-personal and situational changes that mothers with CF encounter in their daily lives. Secondly, further work on the descriptive preliminary theoretical model developed from the analysis of data should be aimed towards continuing to modify and clarify the relationships of the categories that emerged. Specifically, future

work will need to be directed towards testing the theoretical model to reaffirm its richness, depth, and empirical validity.

Summary

It was in the very dailiness, the repetitive dailiness, that the real power of the participants' stories lay. They were not sentimental nor trivial in their stories, but in plain, matter-of-fact, and in the end, unforgettable voices. The participants independently developed strategies for family support and mechanisms to deal with the social control. Regardless of the challenges of child and disease, "living was measured in the doing".

The journeys of the women in this study were thoughtful and real. In their images were yearning, peace, solitude, and togetherness. Through it all, their struggles and triumphs spoke of the monumental significance of simple everyday things. Twentieth century advances in CF care have helped to transform the futures of women in the twenty first century. The participants in this study are the pioneers and roles models for future generations of girls with CF. As mothers, wives, daughters, grandmothers, friends, and neighbors, they strengthened the communities in which they lived, and in turn drew upon the support of those around them. They were empowered by the will to live and by confidence in their destiny.

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Appendix A

Study Instruments:

Demographic Information Form

Letters to Participants and Health Care Providers

Demands of Illness Inventory (DOII)

Self-Management Behavior Form (SMQ-CF)

NIH Clinical Score for CF (NIH-CSCF)

Demographic Information Form

The following information is confidential and is intended to better understand the entire sample of women who respond to the questionnaire.
Please answer the following questions as indicated:

1. What is your age?
    1. Less than 18 years
    2. 19-29 years
    3. 30-39 years
    4. Greater than 40 years

2. What is your level of education?
    1. Less than high school
    2. High school graduate
    3. Some college
    4. College graduate
    5. Graduate school

3. What is your ethnic background?
    1. Caucasian
    2. African-American
    3. Hispanic
    4. Asian
    5. Other

4. What is your religious preference?
    1. Protestant
    2. Catholic
    3. Jewish
    4. Eastern
    5. Other or none

5. Are you currently employed?
    1. Yes
    2. No

6. How long have you worked in your current position?
    1. Less than 6 months
    2. 6 months to 1 year
    3. 1 to 5 years
    4. More than 6 years

7. Which of the following best describes your position in the organization?
    1. Professional
    2. Technical
    3. Clerical
    4. Other

8. How would you describe your current state of health?
    1. Excellent
    2. Good
    3. Fair
    4. Poor

9. How many children do you have?
    1. One
    2. Two
    3. More than two

10. Describe the length of your (last) pregnancy:
    1. Full term
    2. One month early
    3. Two months early
    4. More than two months early

11. Was the pregnancy planned?
    1. Yes
    2. No

12. Were you aware you could get pregnant?
    1. Yes
    2. No

13. Will you or was your child clinically tested for cystic fibrosis?
    1. Yes
    2. No
    3. Do not plan on having tested
    4. Has not been tested yet

14. The test for cystic fibrosis for your child was:
    1. Negative
    2. Positive
    3. NA

15. What is the status of your relationship with your partner?
    1. Married
    2. Single
    3. Divorced
    4. Living together

Letter for Participants

Dear _____, Date of Letter

Enclosed is a copy of the questionnaire for my research. I appreciate your taking the time from your busy lives to complete it.

Your input will help CF health care providers better counsel their patients regarding issues of reproduction. Additionally, your experiences will help young girls with CF have hope for a brighter adulthood. You are truly pioneers for the next generation.

Your responses will be confidential. Please enclose **one** copy of the consent form and the completed questionnaire into the stamped self-addressed envelope. You may keep one copy of the consent form for yourself.

Lastly, if you are interested in the results of the study please be sure to indicate this on the demographics form.

If you have any questions, please feel free to contact me either via Email or by phone.

Sincerely,

Bethany Geldmaker, RN, MSN, PNP,
Phone: 804-XXX-XXXX

Email: Bgeldmaker@HSC.VCU.EDU
Web site: http://home.earthlink.net/~bgeldmaker/

Letter to Health Care Providers

Date of Letter

Bethany Geldmaker
Street Address
Richmond, Virginia
XXXXX
804-XXX-XXXX

To whom it may concern,

_____ indicated she was being followed by your CF Center. She is participating in a study examining outcomes of pregnancy in women with CF. The study has been approved through the Human Subjects Committee of my institution. The women voluntarily respond to be part of the study through contact over the Internet.

A consent for release of medical records information has been signed. I have included a copy of the signed consent for your review. I would appreciate it if you would take 15-20 minutes of your time to complete the enclosed severity of illness measure and mail it back to me in the stamped, self-addressed envelope.

I appreciate your assistance in this matter. Please feel free to contact me with any questions. I look forward to hearing from you.

Sincerely,

Bethany Geldmaker, RN, PNP, MSN
Doctoral Candidate
Medical College of Virginia Campus of
Virginia Commonwealth University

DOII1 Participant Code _____

Demands of Illness Inventory

Below is a list of events and thoughts that describe experiences some individuals have when they experience a health problem. Read the items carefully and determine the extent to which you had these experiences as the result of your cystic fibrosis (CF) **during the last month including today.**
Note: Please mark NA only if the item is not applicable to your particular situation, otherwise mark 0 to 4. Please do not SKIP any items. Thank you! Your answers are confidential and will not be shared with your doctor or nurse unless you request it.

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As the result of my illness I think about:

1. The value my life has for me.	NA	0	1	2	3	4
2.How long I might live.	NA	0	1	2	3	4
3. Not being able to achieve my goals in life.	NA	0	1	2	3	4
4. How I might reorder the priorities in my life.	NA	0	1	2	3	4
5. My own mortality.	NA	0	1	2	3	4
6. How unprepared I've been for this experience.	NA	0	1	2	3	4
7. The uncertainties I face.	NA	0	1	2	3	4
8. If my life will ever return to normal.	NA	0	1	2	3	4
9. What will happen to my family in the future.	NA	0	1	2	3	4
10. Whether my children will face the same illness.	NA	0	1	2	3	4
11. Not having any past experience to relate to this one.	NA	0	1	2	3	4
12.How my experience compares with others having the same or a similar experience.	NA	0	1	2	3	4
13. Why is this happening to me?	NA	0	1	2	3	4
14. How unfair this experience has been.	NA	0	1	2	3	4

15. My odds of getting this illness.	NA	0	1	2	3	4
16. What has caused the illness.	NA	0	1	2	3	4

Page DOII2

Participant Code____

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As the result of my illness our family:

17. Income has gone down.	NA	0	1	2	3	4
18. Doesn't have enough time or energy for recreational activities outside our home.	NA	0	1	2	3	4
19. Doesn't have enough money to support our usual lifestyle.	NA	0	1	2	3	4
20. Doesn't have enough time or energy to entertain friends at home.	NA	0	1	2	3	4
21. Doesn't have enough money for our health care bills.	NA	0	1	2	3	4
22. Doesn't have enough time or energy to go out with friends.	NA	0	1	2	3	4
23. Has had to change our old meal patterns.	NA	0	1	2	3	4
24. Has had to change our child care arrangements.	NA	0	1	2	3	4

As the result of my illness:

25. The children take more responsibility for household tasks.	NA	0	1	2	3	4
26. My partner takes more responsibility for household tasks.	NA	0	1	2	3	4
27. The quality of my sexual activities has changed.	NA	0	1	2	3	4
28. The frequency of my sexual activities has changed.	NA	0	1	2	3	4
29. There isn't time or energy for sexual activities.	NA	0	1	2	3	4
30. I worry about how my children are reacting to my illness.	NA	0	1	2	3	4
31. The children need more emotional support.	NA	0	1	2	3	4

32. I need more emotional support from my family.	NA	0	1	2	3	4
33. The children need more information.	NA	0	1	2	3	4
34. There is a strain on my relationship with my partner.	NA	0	1	2	3	4
35. My partner has had difficulty understanding my feelings.	NA	0	1	2	3	4

Page DOII3

Participant Code _____

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As the result of my illness:

36. I worry about how my partner is responding to my illness.	NA	0	1	2	3	4
37. I wish my partner were handling the illness situation better.	NA	0	1	2	3	4
38. I need to be more sensitive to my partner's moods.	NA	0	1	2	3	4
39. I need to provide more emotional support to my partner.	NA	0	1	2	3	4
40. I need to protect my partner from stress.	NA	0	1	2	3	4
41. I need my partner to be more sensitive to my moods.	NA	0	1	2	3	4
42. I need my partner to help me with my treatments.	NA	0	1	2	3	4
43. My partner has had to change his work patterns.	NA	0	1	2	3	4
44. I'm not able to work at my job.	NA	0	1	2	3	4
45. I've had to miss more time at work than usual.	NA	0	1	2	3	4
46. I'm not able to do my usual amount of work.	NA	0	1	2	3	4
47. I've had trouble finding a job.	NA	0	1	2	3	4

As the result of my illness our family has had to:

48. Make new decisions about running the house.	NA	0	1	2	3	4
49. Revise the rules for the children.	NA	0	1	2	3	4
50. Discuss things concerning the children more.	NA	0	1	2	3	4

51. Decide what is really important to us.	NA	0	1	2	3	4
As the result of my illness:						
52. I go out with my friends less.	NA	0	1	2	3	4
53. My social life has decreased.	NA	0	1	2	3	4
54. I often have to help others understand my illness.	NA	0	1	2	3	4

Page DOII4

Participant Code ____

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As the result of my illness:						
55. It's hard to keep up with my usual pace or routine.	NA	0	1	2	3	4
56. People are overprotective of me.	NA	0	1	2	3	4
57. People are less supportive of me as time goes on.	NA	0	1	2	3	4
58. I find that I need to help others accept my illness.	NA	0	1	2	3	4
59. Others do not really know or understand what I am going through.	NA	0	1	2	3	4
60. Others act differently toward me.	NA	0	1	2	3	4
61. It's hard to plan social activities because I don't know how I'll feel.	NA	0	1	2	3	4
As the result of my illness I:						
62. Feel self-conscious about my body.	NA	0	1	2	3	4
63. Feel less attractive.	NA	0	1	2	3	4
64. Feel dissatisfied with the way I look.	NA	0	1	2	3	4
65. Feel I cannot always rely on my body.	NA	0	1	2	3	4
66. Think about my sexual appeal.	NA	0	1	2	3	4

67. Think about the disfigurement caused by surgery/treatment.	NA	0	1	2	3	4
68. Think about possibly needing to undergo surgery that would result in disfigurement.	NA	0	1	2	3	4
69. Think about the possibility of undergoing surgery to improve my appearance.	NA	0	1	2	3	4
70. Think about not being able to have children.	NA	0	1	2	3	4
71. Feel more susceptible to other illnesses.	NA	0	1	2	3	4

Page DOII5

Participant Code _____

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As the result of my illness I:

72. Concentrate on new bodily sensations that may indicate illness.	NA	0	1	2	3	4
73. Worry my illness may reoccur with more severity.	NA	0	1	2	3	4
74. Tend to be preoccupied with the symptoms of my illness.	NA	0	1	2	3	4
75. Think about how I'm handling my illness situation.	NA	0	1	2	3	4
77. Wonder if the illness is spreading undetected.	NA	0	1	2	3	4
78. Wonder why I still receive treatments even though my symptoms have subsided.	NA	0	1	2	3	4
79. Think about the illness being unending.	NA	0	1	2	3	4
80. Worry my health will get progressively worse.	NA	0	1	2	3	4
81. Worry the illness will involve other parts of my body in the future.	NA	0	1	2	3	4

As the result of my medical treatment:

82. I find it difficult to continue with follow-up appointments.	NA	0	1	2	3	4
83. I find it difficult to continue to continue the treatments.	NA	0	1	2	3	4

84. I sometimes think the adverse effects of treatment outweigh the possible benefits. NA 0 1 2 3 4

85. I worry about the expense of treatment. NA 0 1 2 3 4

86. I've changed my diet. NA 0 1 2 3 4

87. I'm more regimented in the time I eat. NA 0 1 2 3 4

88. My whole life is more regimented. NA 0 1 2 3 4

89. I've adjusted the way I exercise. NA 0 1 2 3 4

90. I'm considering the need to undergo more treatment. NA 0 1 2 3 4

Participant Code _____

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As the result of my medical treatment:

91. I'm considering if I should try a different treatment. NA 0 1 2 3 4

92. It's difficult waiting for the results of my medical tests. NA 0 1 2 3 4

93. I worry about the physical side effects of treatment. NA 0 1 2 3 4

94. I worry I'll develop new physical symptoms in the future. NA 0 1 2 3 4

95. I often feel worse rather than better after treatment. NA 0 1 2 3 4

96. It's difficult waiting to undergo treatment or surgery. NA 0 1 2 3 4

At times, my health care providers:

97. Are not sensitive to my preference for treatment. NA 0 1 2 3 4

98. Act as if my opinions are unimportant. NA 0 1 2 3 4

99. Make decisions without my best interests in mind. NA 0 1 2 3 4

100. Do not tell me the truth about my health status. NA 0 1 2 3 4

101. Do not show concern for me as a person. NA 0 1 2 3 4

As I've experienced my illness situation:

102. I do not want my health providers to tell me the truth if I take a turn for the worse.	NA	0	1	2	3	4
103. I want more facts about the treatments.	NA	0	1	2	3	4
104. I have questions that I want to ask but just can't.	NA	0	1	2	3	4
105. I feel rushed to make a hasty treatment decision.	NA	0	1	2	3	4
106. I want to be more assertive about expressing the direction my treatment should take.	NA	0	1	2	3	4
107. I want to be told the reason why, when asked to do something for treatment.	NA	0	1	2	3	4
108. I sometimes don't understand the treatment I'm receiving.	NA	0	1	2	3	4

Page DOII7

Participant Code _____

0=Not at all
1=A little bit
2=Moderately
3=Quite a bit
4=Extremely

As I've experienced my illness situation:

109. I'm satisfied with my hospital care.	NA	0	1	2	3	4
110. I feel my illness is being correctly managed.	NA	0	1	2	3	4
111. I'm confident my health will be correctly managed in the future.	NA	0	1	2	3	4
112. I'm satisfied with the progress of my treatment.	NA	0	1	2	3	4

Self-Management Behaviors in Cystic Fibrosis

Answer the following questions about your medical Problems with CF. **Circle** the answer closest to your choice.

113. How many lower respiratory infections have you had in the past 6 months (bronchitis/pneumonia)? None 1-2 3-4 5-6 >6 Unsure

114. How long has it been since your last lower respiratory infection?	Never	<2 had	2-6 months	7-12 months	>1 months	Unsure yr
115. Have you had problems with digestion (loose fatty stools, stools containing undigested food)?	Yes			No		Unsure
116. Have you had problems with weight gain or growth?	Yes			No		Unsure
117. How long has it been since your last digestion problem you thought was due to malabsorption with CF?	Never	< 2 had	2-6 mos	7-12 mos	> 1 mos	Unsure yr

Answer these questions about what you have been recommended to do at home for your CF. **Circle** the answer closest to your choice.

118. Do you use respiratory therapy as a routine practice (aerosol/inhalation)?	Yes	If yes # times/day_____		No	Unsure
119. Do you use chest physical therapy as routine practice?	Yes	If yes # times/day_____	No	Unsure	

SMQ-CF8 Participant Code_____

120. Do you other kinds of chest physical therapy in addition to or instead of clapping/postural drainage?	Exercise	Yes	No	Unsure
	PEP Device	Yes	No	Unsure
	Mechanical clapper	Yes	No	Unsure
	Other	Yes	No	Unsure
121. Do you have specific diet guidelines for CF?		Yes	No	Unsure
122. Do you have to use high calorie supplements to gain weight?		Yes	No	Unsure

This next section of the questionnaire is interested in finding out the things you do to treat CF. With each question, think about what you have done for each therapy or medical problem. If you have no involvement for an activity, indicate that as **never**. If the section or question does not apply **check not prescribed**.

Read each item carefully and choose the answer that **best** describes your home practices. Most people cannot do all of the things expected of them all the time. Try to be honest as possible about what you really do. Your answers are confidential and will not be shared with your doctor or nurse unless you request it.

0=Never
1=Rarely
2=Sometimes
3=Usually
4=Always

In the past 1 **month**, how have you watched for lower respiratory infection (pneumonia/bronchitis)?

123. Observed how fast breathing at rest as a sign of respiratory infection?		0	1	2	3	4
124. Watched for changes in activity (energy level) in exercise, play, or work?		0	1	2	3	4
125. Watched for changes in hunger/appetite as a sign of respiratory infection?		0	1	2	3	4
126. Experienced changes in mood/temper?	0	1	2	3	4	
127. Watched for weight loss as a sign of respiratory infection?		0	1	2	3	4
128. Watched for decreased food intake as as a sign of respiratory infection?		0	1	2	3	4
129. Watched for increased cough?		0	1	2	3	4
130. Watched for increased sputum/mucus?		0	1	2	3	4
131. Watched for change in the color or thickness of mucus as a sign of respiratory infection?		0	1	2	3	4

Participant Code_____

Think about how you treat a lower respiratory infection. **Circle** how frequently you do each action. Note: If you have not had a lower respiratory infection (bronchitis/pneumonia), **skip** to question **140.**

0=not prescribed
1=Never
2=Rarely
3=Sometimes
4=Usually
5=Always

132. Begin respiratory therapy (aerosol. inhalation) or increase the number of respiratory treatments done each day?	0	1	2	3	4	5
133. Begin chest physical therapy (CPT) or increase the number of CPT sessions done each day?	0	1	2	3	4	5
134. Begin prescribed antibiotics or increase number of antibiotics used with the advice of your doctor's office?	0	1	2	3	4	5
135. Arrange daily time schedule to fit in extra therapy?	0	1	2	3	4	5
136. Contact the doctor's office if no improvement occurs after the change in therapy?	0	1	2	3	4	5
137. Stop treatment at the end of a prescribed course if the symptoms have not disappeared?	0	1	2	3	4	5

Think about the last time you were treated for a lower respiratory infection. **Circle** how often you did each item.

Note: If you have not had a lower respiratory infection (bronchitis/pneumonia) **skip** to question **140.**

138. How long did you observe symptoms before treatment?	1-3 days	4-6 days	7-13 days	2-4 weeks	>1 month
139. How long did you wait after beginning treatment to decide if there was improvement?	1-3 days	4-6 days	7-13 days	2-4 week	>1 month

SMQ-CF10 Participant Code_____

Think about the past month when you did chest physical therapy (CPT). **Circle** how often did you do each of the following actions. Note: if not prescribed, **skip** to question **145.**

0=Not prescribed
1=Never
2=Rarely
3=Sometimes
4=Usually
5=Always
6=Did not occur

140. Coughed intentionally to bring up mucus as a past of CPT?	0	1	2	3	4	5	6
141. Observed the amount of mucus to decide if CPT was effective?	0	1	2	3	4	5	6
142. Observed cough to decide if CPT was effective?	0	1	2	3	4	5	6
143. When unable to do CPT, made arrangements for someone else to do it ?0		1	2	3	4	5	6
144. Missed how many CPT therapies during the last week used?		Not prescribed	None	Almost none	1/4	½	3/4 or more

Think about the **last week** you used respiratory therapy (RT=aerosol/inhalation). **Circle** how often you did each action. Note: If not prescribed, **skip** to question **149.**

145. Observed cough to decide if RT was effective?	0	1	2	3	4	5	6
146. Observed mucus production to decide if RT was effective?	0	1	2	3	4	5	6
147. Increased the number of RT treatments with an increase in symptoms?	0	1	2	3	4	5	6
148. Missed how many RT treatments during the last week used?	Not prescribed		None	Almost none	1/4	½ or more	3/4

SMQ-CF11 Participant Code_____

In the past 1 **month**, how have you watched for problems with digestion (malabsorption). Circle how often you did each action. Note: If you do not have malabsorption (pancreatic insufficiency) **skip** to question **160.**

0=Never
1=Rarely
2=Sometimes
3=Usually
4=Always

149. Looked at stools (bowel movements) for changes at least once a week?	0	1	2	3	4
150. Noticed the number of stools in a day?	0	1	2	3	4

151. Watched for greasy changes in stools as a sign of poor digestion?	0	1	2	3	4
152. Watched for floating of stools as a sign of poor digestion?	0	1	2	3	4
153. Observed for stomach aches or cramps as a sign of poor digestion?	0	1	2	3	4
154. Watched for stomach bloating or increased gas as signs of poor digestion?	0	1	2	3	4
155. Observed change in appetite as a sign of poor digestion?	0	1	2	3	4

Think about how you treat poor digestion (malabsorption). **Circle** how often you do each action. Note: If you do not have malabsorption, **skip** to question **160.**

0=Not prescribed
1=Never
2=Rarely
3=Sometimes
4=Usually

156. Adjust dose of pancreatic enzymes depending on the amount of food eaten?	0	1	2	3	4
157. Adjust dose of pancreatic enzymes depending on type of food eaten (like fruit/meat/dairy product)?	0	1	2	3	4
158. Take enzymes before or during meals of snacks?	0	1	2	3	4
159. Make enzymes available to use with meals or snacks eaten away from home?	0	1	2	3	4

Participant Code_____

Think about your diet and eating behavior over the past 1 **month. Circle** how often you did each action.

0=Never
1=Rarely
2=Sometimes
3=Usually
4=Always

160. Observed food intake?	0	1	2	3	4

161. Observed body appearance for signs of weight loss?	0	1	2	3	4
162. Noted fit of clothes as a sign of weight loss or gain?	0	1	2	3	4
163. Went over growth/weight trend (growth chart) with a member of the health care team?	0	1	2	3	4
164. Used high calorie snacks to add extra calories to diet?	Not Prescribed 0	1	2	3	4
165. Added more salt to diet for increased sweating (as in warm weather)?	Did not occur 0	1	2	3	4
166. Increased calorie intake when weight loss or poor growth was noticed?	Did not occur 0	1	2	3	4
167. Discussed poor weight gain with a member of the health care team?	Did not occur0	1	2	3	4
168. Missed how many doses of vitamins in the past week?	Not prescribed	None	Almost none	1/4	½3/4 or more

As a result of my pregnancy:

169. I seek more immediate attention for illness symptoms.	0	1	2	3	4
170. I find I have less time to do my therapies.	0	1	2	3	4
171. I feel I have achieved something I never thought possible.	0	1	2	3	4
172. I set goals for myself.	0	1	2	3	4
173. The future looks brighter.	0	1	2	3	4
174. Life has more meaning.	0	1	2	3	4

NIH Clinical Scoring for Cystic Fibrosis

Name_____ Date of Birth_____ Study Code#_____
Date of Rating_____ Name of Rater_____

	Point Range	MAX	Score
1. **Pulmonary**			
A. **X-ray**		**17**	
1. Minimal accentuation of pulmonary markings:	1 , 2, 3		
2. Increased pulmonary markings: mild overaeration; atelectesis; and/or mucus plugging:	4, 5, 6		
3. Moderate overaeration: fibrosis;atelectesis; and/ or mucus plugging; early cyst formation:	7, 8, 9, 10		
4. Severe overaeration; extensive fibrosis and cyst formation; pulmonary obstruction; bronchiectesis:	11. 12, 13		_____
5. Acute infiltrate:	1, 2, 3. 4		_____
B. Pulmonary Function Tests		**17**	
1. Forced Vital Capacity			
less than 90% of predicted	1		
less than 80% of predicted	3		
less than 70% of predicted	5		
less than 60% of predicted	7		
less than 50% of predicted	9		_____
2. FEV1			
less than 70% of predicted VC	1		
less than 66% of predicted VC	2		
less than 58% of predicted VC	4		
less than 50% of predicted VC	6		
less than 42% of predicted VC	8		_____
C. Pulmonary exacerbation requiring intensive therapy		**5**	
1. Past 3 months	5		
2. Past year	3		_____
D. Pneumotborax		**5**	
1. Past 6 months or recurrent	5		
2. Ever	3		_____
SUBTOTAL PULMONARY PAGE 1			_____

NIH-CF2

Participant Code____

	Point Range	MAX	Score
Pulmonary Continued:			
E. Hemoptysis (omit if none since pulmonary surgery)		7	
1. Massive			
a. Past 6 months	7		
b. More than 6 months ago	4		
2. Small amount in past year			
a. Rare streaks: with acute infection or < 15 ml	1		
b. occasional streaks or less than 30 ml	2		
c. frequent streaks or greater than 30 ml	3		____
F. Pulmonary Surgery		7	
1. Amount of lung resected			
a. Segment	1		
b. Lobe	2		
c. Right lung 2 lobes	3		
d. Right or left lung	4		____
2. Length of time since surgery			
a. ≥ 5 years	1		
b. 1-5 years	2		
c. 0-12 months	3		
G. Cor Pulmonale (based on symptom state in last month)		5	
1. Asymptomatic	3		
2. Symptomatic- controlled with therapy	4		
3. Symptomatic- intractable despite therapy	5		

SUBTOTAL PULMONARY PAGE 2 _____

NIH-CF3

Participant Code_____

	Point Range	MAX	Score
Pulmonary Continued:			
H. Physical examination (based on last 1 month)		9	
1. Clubbing			
a. Mild	1		
b. Moderate/Severe	2		
2. Chest deformity (A-P diameter, rib flare, kyphosis)			
a. Mild	1		
b. Moderate	2		
c. Severe	3		
3. Cyanosis present	1		
4. Breath Sounds			
a. Crackles/wheezes- isolated	1		
b. Crackles/wheezes- multilobe	2		
c. Poor airflow	3		_____
I. Sputum Production and/or Cough (based on last 1 month)		3	
1. Cough alone (dry cough w/CPT or activity)	1		
2. Sputum production (wet cough) with pulmonary exacerbation	2		
3. Chronic sputum production (chronic cough wet or dry)	3		_____

Subtotal Pulmonary Page 3	_____
Subtotal Pulmonary Page 2	_____
Subtotal Pulmonary Page 1	_____
TOTAL PULMONARY	_____

NIH-CF4 Participant Code_____

	Point Range	MAX	Score
II. **GENERAL**			
A. Weigbt		**5**	
1. Poor appetite (in past 6 months)			
a. Occasional with pulmonary exacerbation	1		
b. Chronic/ frequent	2		
2. Less than third percentile	2		
3. Loss of more than 2 kg past 3 months	2		
4. Loss of more than 5 kg past year or significant decline from growth curve	3		_____
B. Activity		**11**	
1. Energy (in last 1 month)			
a. Less energetic- tired at end of the day (than same age peers)	1		
b. Moderate limitation- cannot go through day without rest (needs more than peers but continues active play/movement)	2		
c. Marked limitation- requires multiple rest periods during the day (restricted to quiet play/rest with little or no active movement)	3		
2. School/work (in past 6 months or last semester) (If not in school/work use number of days not able			

to participate in daily play, activities, or daycare.

a. Fair attendance (absence 5-15 days)	1	
b. Poor attendance (absence > 15 days)	2	
c. Homebound (marked motor restriction)	3	_____

SUBTOTAL GENERAL PAGE 4 _____

NIH-CF5

Participant Code_____

	Point Range	MAX	Score
General Continued:			
3. Exercise tolerance (in last 1 month)			
a. Slight limitation or sustains strenuous exercise (keeps or sustains movement usually but slower)	1		
b. Tires during exertion, rests voluntarily (in play/ movement)	2		
c. Limits exercise (movement) due to dyspnea (can't keep up with peers/maintain sustained movement of limbs or head erection)	3		
d. Dyspnea with minimal exertion	4		
e. Dyspnea at rest	5		_____
C. Attitude		**9**	
1. Compliance			
a. Clinic Attendance (in past 6 months)			
1.) Partial/forgot or missed follow-up appointment	1		
2.) Poor/forgot or missed more than 2 appointments	2		

b. Therapy Prescribed (last 1 month)

1.) CPT/respiratory therapy

a.) Partial/irregular (lapses up to 2 weeks) 1

b.) Poor- rarely performs (lapses > 2 weeks) 2

2.) Medication

a.) Partial/irregular (missed up to 1 week) 1

b.) Poor (missed > 1 week) 2 _____

SUBTOTAL GENERAL PAGE 5 _____

NIH-CF6

Participant Code_____

	Point Range	MAX	Score

General Continued:

3. Depressed philosophy of CF (based on state in last 1 month)

a.) Feelings of helplessness, depression, loss of control 1

b.) More severe depressive or loss of control, problems with some dysfunction 2

c.) Disabled functioning due to loss of control, depression, and/or denial 3 _____

SUBTOTAL GENERAL PAGE 6 _____

SUBTOTAL GENERAL PAGE 5 _____

SUBTOTAL GENERAL PAGE 4_____

TOTAL GENERAL _____

POINTS DEDUCTED

PULMONARY _____

GENERAL _____

TOTAL POINTS DEDUCTED _____
MAX=100

Final Score 100 - total points deducted = _____

Appendix B

Copy of Web Site
Adult CF Newsletter Abstract

Bethany Geldmaker,
RN, MSN, PNP
Doctoral Student

OUTCOMES OF PREGNANCY IN WOMEN WITH CYSTIC FIBROSIS

OUTCOMES OF PREGNANCY IN WOMENWITH CYSTIC

Information About The Study

Information About Myself

Directory of Related Links

Information I Need To Contact You

I am interested in talking with women who have cystic fibrosis and who are pregnant or who have children.

Your experiences are important to younger generation of girls who are planning for their futures. They are also important to the staff who provide your medical care.

Page 2 of my home page provides more detail about the research study. Page 3 gives you background about myself. Page 5 enables you to contact me to participate in my study.

Please feel free to contact me with any questions. I look forward to hearing from you!

Medical College of
Virginia Campus of
Virginia Commonwealth
University
Richmond, VA, USA

Research Abstract

The increase in life expectancy from adolescence to young adulthood, for women with cystic fibrosis, allows them to consider becoming pregnant and having children. The purpose of this research is to examine what demands the chronic illness of cystic fibrosis places on women during and after pregnancy.

The sample for the study is women with cystic fibrosis who are pregnant or have children and who agree to participate in the study.

There are 3 parts to this study. Women who would like to participate in the study are asked to fill out a questionnaire that asks about how they manage their cystic fibrosis in their personal lives and about the kinds of care their illness requires at home. The questionnaire takes about 25 minutes to complete and is available only in English.

The second part of the study is to talk with myself about ways the kinds of experiences they have had during their pregnancy and in caring for their families. The third part of the study is information about pulmonary function studies.

If you would like to be part of my study, please complete the information on page 5 so I may contact you.

My Professional Background

My Personal Background

Directory of Related Links

OUTCOMES OF PREGNANCY IN WOMEN WITH CYSTIC

Information About The Study

Information About Myself

Directory of Related Links

Information I Need To Contact You

Below I have listed several other web sites. You may wish to explore these if you wish additional information about cystic fibrosis.

Happy exploring!

Web site name and address hyperlink
Briefly summarize why the information on the site to which you're linking is useful.

Web site name and address hyperlink
Briefly summarize why the information on the site to which you're linking is useful.

Bethany Geldmaker,
RN, MSN, PNP

Information I Need To Contact You

OUTCOMES OF PREGNANCY IN WOMEN WITH CYSTIC

Information About The Study

Information About Myself

Directory of Related Links

Information I Need To Contact You

If you wish to participate

Please provide the following information in the spaces provided below : name, mailing address, Email address, and phone number with area code.

Comments:

Address:

City: State/Prov:

Country: Zip/Post. code:

Phone:

E-mail:

Adult CF Newsletter Abstract

RESEARCH STUDY ON MOTHERS AND PREGNANT WOMEN WHO HAVE CYSTIC FIBROSIS:

Editor's Note: *Bethany Geldmaker is a doctoral student at Virginia Commonwealth University who has worked as a nurse practitioner in Pediatric Pulmonary Medicine with children and adults with CF. She has nursing experience working with mothers and children.*

During this employment Bethany became aware of the lack of information available about how women deal with their pregnancy, children, and disease. What follows is an abstract for the research she would like to pursue in response to her aforementioned observations.

ABSTRACT:

The increase in life expectancy from adolescence to adulthood, for women with CF, allows them to consider becoming pregnant and having children. The purpose of this research is to examine what demands the chronic illness of CF places on women during and after pregnancy. The study is designed to examine the experience from the perspective of the women.

The sample for the study consists of women with CF who are pregnant or have children, agree to participate in the study, and who speak English.

There are 3 parts to the study. Women who would like to participate in the study are asked to fill out a questionnaire that asks about how they manage their CF in their personal lives and about the kind of care their illness requires at home. The questionnaire takes about 25 minutes to fill out and is available only in English.

The second part of the study is to talk with myself about the kinds of experiences they have had during pregnancy and in caring for their families. The third part of the study is information about their pulmonary function studies.

If you have CF, are either a mother or currently pregnant, and wish to participate in this study, Bethany would welcome your enquiry. She can be contacted via one of the following:

Web-Site: **http://home.earthlink.net/~bgeldmaker/**

E-Mail: **BGeldmaker@hsc.vcu.edu**

Telephone: [redacted]

Bethany Geldmaker RN, PNP, PhD (c)

Appendix C

Content Outline for Participant Interviews

Content Outline for Participant Interviews

The following questions will serve as the template for interviews:

1. Questions regarding the participants's pregnancy:

    1. What things did you think about when deciding to get pregnant (or keep the baby)?
    2. What was the response of people when you told them you were pregnant?
    3. Who was the first person you told? Why?
    4. What was the most rewarding experience about being pregnant?
    5. What was the most challenging experience about being pregnant?
    6. Tell me about a situation you found difficult about providing care for yourself during pregnancy and what you did to manage the situation?

2. Questions related to issues of child care:

    1. What did you find most rewarding in caring for the baby?
    2. What do you find most challenging about caring for the baby?
    3. Tell me about a situation you found difficult about providing care to your child, describe what you did and what were your thoughts.
    4. What has helped you the most in adjusting to caring for the baby?
    5. Tell me your top 5 concerns in dealing with caring for yourself and your child.
    6. How well do you feel you are dealing with care of yourself and your family?
    7. What would help you do better? What helps you the least?
    8. What do you do when you are sick or need to be hospitalized?
    9. What is the hardest part of having CF and a child?
    10. Who has helped you the most in dealing with your CF and your child; in what way have they been helpful?
    11. In what way has being pregnant and having a child changed you?

Appendix D

Informed Consent Form

Consent to Participate in a Research Study
Outcomes of Pregnancy in Women with Cystic Fibrosis

Principle Investigator
Bethany Geldmaker, RN, M.S.N., P.N.P.
Medical College of Virginia Campus of Virginia Commonwealth University

Dissertation Advisor
Rita H. Pickler, RN, Ph.D.
Medical College of Virginia Campus of Virginia Commonwealth University

Introduction

I am interested in finding out from you what it is (or was) like for you to have CF and be pregnant. My study has two parts: one part is a questionnaire and the second part is an interview. I am specifically interested in how you were able to take care of yourself, your disease, and your pregnancy. In addition to your pregnancy, if you have a child, what is it like to care for yourself, your disease, and for your child? There are 2 questionnaires that ask you to rate these experiences. If you chose to participate, it will take about 30 minutes of your time to fill them out. I would also like permission to obtain data from your chart including lung function tests (PFTs), sputum cultures, and your height and weights.

For the second part of my study, I would like to contact you and talk to you about what is like to be pregnant, care for yourself, and now care for your child. There may be things the questionnaire does not ask that would be important for me to know. I would like permission to contact you to ask questions about your experiences. Would you be interested in being contacted? Yes___ No ___. The interviews will be audio taped.

Benefits

There are no personal benefits to your participation in this study. However, information learned from your participation maybe beneficial to assist other women with CF who are considering pregnancy.

Alternative Therapy
This is not a therapeutic study. You have the alternative to not participate in the study.

Risks, Inconveniences, Discomforts
There are no risks involved in this study.

Initials_____

Costs of Participation

There are no costs related to your participation in this study. You will not be paid for participating in this study.

Confidentiality of Records

The investigator will treat your identity with professional standards of confidentiality. The information obtained in this study may be published, but your identity will not be revealed. The audio tapes from interviews will be destroyed at the end of the study.

Withdrawal

Participation in this study is voluntary. The investigator will answer any questions you may have about the study. You are free to withdraw your consent and discontinue participation at any time. If you decide to withdraw or have any questions, please call Bethany Geldmaker at (804) 780-8243 (this is voice mail--please leave a message) or Rita Pickler at (804) 828-0721. Discontinuation in this study will in no way affect or jeopardize the care you receive now or in the future at this institution.

Subjects' Rights Information

If you have any questions about your rights as a research subject, you may contact the Committee on the Conduct of Human Research at (804) 828-0868 for information or assistance.

You will receive a copy of this consent form.

_____ _____

Signature Date

_____ _____

Investigator Signature Date

Vita