

CHRONIC SORROW AND ILLNESS AMBIGUITY IN CAREGIVERS OF
CHILDREN WITH SICKLE CELL DISEASE

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A THESIS

Submitted to
Michigan State University
in partial fulfillment of the requirements
for the degree of

Human Development and Family Studies—Master of Science

2013

ABSTRACT

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The purpose of this study was to give voice to primary caregivers of children with sickle cell disease. The research was supported through use of the FAAR Model, Life Course Theory, and Ambiguous Loss Theory. Two caregivers participated in a semi-structured interview to tell their story with sickle cell disease. The caregivers noted common themes including knowledge of the disease, family routine and roles, and resources for support. The caregivers presented a contrasting experience which highlighted how illness can affect the child, the family, and the family's adaptation to illness.

This thesis is dedicated to the patients and families I have had the privilege of caring for over the years. They are a true inspiration.

ACKNOWLEDGEMENTS

I want to thank the caregivers at Hurley Medical Center who were willing to share their story with sickle cell disease. I was granted a brief glimpse into their family life to better understand how to help other families. Thank you to Dr. Ames for being my adviser and committee chairperson. Your guidance ensured I would navigate the journey towards my Masters degree. Thank you to Dr. Carolan and Dr. Onaga for recognizing my passion in supporting families during stressful times. The utmost of thanks goes to my family for encouraging me along the way. This thesis has been a labor of love and they have guided me when I became discouraged. I love you all. Finally, thank you Dad for watching over me from above.

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CHAPTER ONE

INTRODUCTION

BACKGROUND OF THE PROBLEM

Sickle cell disease affects one in every 500 African Americans in the United States (Mitchell, Lemanek, Palermo, Crosby, Nichols, & Powers, 2007). Advancing medical technologies are increasing survival rates for children with sickle cell disease. Yet, there continues to be a lack of awareness surrounding the continual burden illness places on primary caregivers. Chronic sorrow has been highlighted as an area of concern for ineffective coping for mothers of children with sickle cell disease. Families must now become experts in their child's care as well as deal with the emotional toll of chronic illness on the family. Primary caregivers of children with sickle cell disease also may have feelings of guilt regarding the genetic component of the disease. Family systems must adapt as members begin to accommodate the ambiguity of sickle cell disease (Northington, 2000). Chronic sorrow affects caregiver coping, especially during times of acute hospitalization, illness progression, and missed developmental milestones. Chronic sorrow is exacerbated by the pile up of stressors and the complexity of illness related ambiguous loss (Melnik, Feinstein, Moldenhouer, & Small, 2001).

Chronic sorrow has been studied in caregivers across a variety of acute and chronic illnesses, yet there continues to be lack of replicated data on specific populations. To date, there is only one published study regarding the feelings of chronic sorrow in African American parents of children with sickle cell disease (Northington, 2000). The diagnosis of sickle cell disease brings a level of uncertainty regarding disease management and outcome for each child. Upon diagnosis of a chronic illness, parents begin to grieve the loss of the perfect child. Denial of the diagnosis or the pervasiveness of the disease is common, but often does not continue because

management of the illness demands parents begin coping (Melnik, Feinstein, Moldenhouer, & Small, 2001). The initial grief of diagnosis and the genetic component of sickle cell disease may be overwhelming. Parents must begin to educate themselves on their child's illness and will begin to incorporate disease management into family life. At the time of diagnosis, parents are taught to monitor their child for any signs of sickle cell crisis that may require medical attention (Northington, 2000). Complications associated with sickle cell disease may include dehydration, pain episodes, stroke, infection, and delayed development. The constant need for monitoring can severely affect the parent's ability to cope and hinder family adaptation (Ievers, Brown, Lambert, Hsu, & Eckman, 1998). Parents may have to adjust their parenting practices and take on new roles as medical caretakers. Family functioning must adapt to accommodate daily medical care, doctor appointments, and increased financial stress (Melnik, Feinstein, Moldenhouer, & Small, 2001).

A parent's ability to cope well and manage feelings of chronic sorrow is largely dependent upon the family's ability to adjust and adapt to the child's illness. The FAAR model shows the need for balance between the stressors and the resources available to manage such stressors (McCubbin & Patterson, 1983). The demands and stressors of a child with a chronic illness are high and can be intensified with hospitalizations and medical complications (Patterson, Holm, & Gurney, 2004). The cumulative effect of illness related family and daily life stressors make family coping and adaptation seem impossible. Family resiliency decreases as the pile up of stressors overwhelms the family system (Boss, 2006). Previous studies of chronic sorrow have indicated parental feelings of lack of resources and support from the healthcare team. Interventions aimed at providing support and enhancing coping skills would be beneficial

if triggers of chronic sorrow for parents of children with sickle cell disease could be identified (Northington, 2000).

Despite medical advances, children with sickle cell disease still have an unclear prognosis and may have a limited life expectancy. The ambiguity of chronic illness can make it difficult for parents to cope effectively and function well as a family (Berge & Holm, 2007). Mastery of a chronic illness is almost impossible because of the ambiguity. Meaning making is an essential part of family coping and adaptation. During episodes of chronic sorrow, parents may have to search for new meaning, and find hope within the unknown. Each family member must find meaning with the chronic illness in order for the whole family system to move forward (Boss, 1999).

When a child is initially diagnosed with sickle cell disease, the primary caregiver begins to evaluate existing stressors and resources available for family support. Despite medical advances, the outcomes for children with sickle cell disease are still unclear. Caregivers may experience feelings of ambiguous loss as they try to grasp the future outcomes for their child. Feelings of loss are amplified by existing stressors and reduced by resources available for parent support. Feelings of ambiguous loss can amplify into a recurrent grief known as chronic sorrow. Chronic sorrow is continuous but managed through family adaptation, positive coping strategies, and healthcare team support.

Hurley Medical Center is a 443 bed teaching hospital serving the residents of the city of Flint, Michigan and greater Genesee County (Hurley Medical Center, 2011). African Americans comprise 53.27% of the population in the city of Flint (The Health of Genesee County, 2008). High poverty rates and low median incomes can affect health outcomes for many of the patients at Hurley Medical Center. Current statistics indicate that 1 in every 500 African Americans have

sickle cell disease, making Hurley Medical Center a primary resource for health care (Mitchell, Lemanek, Palermo, Crosby, Nichols, & Powers, 2007).

PURPOSE OF THE STUDY

The purpose of this study is to give voice to primary caregivers of children with sickle cell disease. It is still not fully understood how individual families adapt and function while managing the burden of their child's medical care. Family centered care initiatives are pushing medical professionals to provide better social and emotional support to children and their caregivers. Through research like this study, the healthcare team can be better informed to provide support during times of optimal health and during times of medical crisis. Concerns with lack of medical adherence may be better understood through examination of parent resources and supports. This case study research will help gather valuable information regarding how caregivers manage feelings of ambiguous loss and recurrent feelings of chronic sorrow.

DEFINITIONS

For the purpose of this study, the following definitions are being used:

- Primary caregiver: the primary person in charge of the child's daily care and well being.
- Sickle cell disease will include those children with sickle cell anemia (SS), sickle-hemoglobin-c disease (SC), sickle beta-plus thalassemia, and sickle beta-zero thalassemia (The Management of Sickle Cell Disease, 2002).
- Chronic sorrow: any recurrent feelings similar to feelings experienced at the time of the child's initial diagnosis.
- Family adaptation: any reorganization of roles within the family and change in behaviors to optimize family functioning post diagnosis.

- Resources: sources of support that buffer the effects of crisis on the family.
- Stressors: events or transitions that force change within the family system.
- Emotions surrounding illness: positive and negative emotions associated with illness or overall family change.
- Coping skills: strategies used by the caregiver to decrease stressors and manage the need for continual family adaptation.

RESEARCH QUESTIONS

This study aims to investigate:

- (1) How is chronic sorrow a factor in daily family functioning?
- (2) How do caregivers of children with sickle cell disease cope with the unknown future?
- (3) How do caregivers describe their experience with the medical team?

THEORETICAL FRAMEWORK

This research will be developed through use of three theoretical perspectives. The FAAR Model helps to examine how stressors and resources can help a family adapt to change. The Life Course Theory will represent how family members and the family unit change over time. Finally Ambiguous Loss Theory helps explain the ambiguity in unresolved loss and the grief associated with chronic illness.

FAAR MODEL

The FAAR Model was developed through use of Hill's ABCX framework and McCubbin and Patterson's Double ABCX Model. The Hill ABCX framework examined how stressors (A), resources (B), and meaning of the event (C) interact to produce the final outcome for the family (X). Stressors and Hardships are the (A) in Hill's model. The stressor could be an event or change in family life. The hardship is a demand placed upon the family because of the stressor.

The resources (B) encompass what the family has or uses to overcome the stressor and hardship. The resources can be used to buffer the cost of stress on the family. The meaning of the event (C) helps the family define whether there is a crisis and change needed. Family values and beliefs help attach meaning to an event. Finally, family crisis (X) occurs when the stressors, hardships, resources, and meaning of the event create the need for family adaptation (McCubbin & Patterson, 1983).

The ABCX Model was used by researchers to analyze the stressors, resources, and outcomes of families of Vietnam veterans participating in a longitudinal study. The study gave insight that there were additional factors missing from the ABCX Model that appeared to affect the family's ability to adapt. The Double ABCX Model noted factors that affected the family post crisis. After reaching crisis state, some families experienced a pile up of additional stressors. New resources and social supports were developed within the extended family and community. Some families were able to redefine the meaning of the crisis. These families decreased the emotional intensity of the crisis and sought the experience as a time for family growth. New coping strategies were developed to decrease stressors and increase resources. These coping strategies affected the meaning attached to the crisis and ultimately could change the ability for adaptation in the family. Adaptation for the family occurs after members seek to find a new sense of balance within their family and within their community. This sense of balance is aided with family coherence. The family members must find and accept a new sense of normal which results in positive family adaptation (McCubbin & Patterson, 1983).

The FAAR Model was developed through additional research that integrated the Double ABCX Model and processes of which variables interact. The FAAR Model shows the need for balance between the stressors and the resources available to manage such stressors. The model

involves a phase of adjustment and a phase of adaptation. During the adjustment phase, the family may resist making family changes or ignore the demands of the stressor. These coping strategies are only temporary and are used to minimize the reality of the impending crisis. At some point, family members begin to restructure their roles to meet new demands and maximize resources. The family also redefines the stressor and decides whether adaptation is possible. Coping strategies are developed and modified to meet new stressors. Adaptation can have positive or negative effects on the family. Many families cycle through adjustment and adaptation several times as new stressors arise, resources decrease, or family meaning changes (McCubbin & Patterson, 1983).

LIFE COURSE THEORY

Life Course Theory aims to study how family members and the family as a whole change over time. This theory emphasizes five major themes. First, family change is affected by time. This time includes changes because of ontogenetic time, generational time, and historical time. Secondly, the social context in which the family lives is very important. This may include the micro and macro levels of their environment as well as cultural and social changes. The meaning attached to events also affects the family's social context. Third, this theory focuses on process and change and how family member behaviors affect the change over time. Fourth, this theory addresses typical and atypical behaviors over time. Atypical behaviors represent diversity in family members. During certain life stages, diversity may be more typical than atypical. Finally, Life Course Theory encourages multidisciplinary analysis (Bengtson & Allen, 1993).

AMBIGUOUS LOSS THEORY

Ambiguous loss theory was developed by researcher Pauline Boss after studying families in therapy. Boss (2007) noted that while loss is a universal experience many families struggled

with loss experiences involving ambiguity (Boss, 2007). A loss that is not finalized creates confusion within the family. The family may be uncertain about the need to change family roles and rules. Ambiguity may arise from a family member being physically present, but psychologically absent such as individuals with Alzheimer's disease or other mental impairment. Family members might also be psychologically present, but physically absent such as missing children or soldiers missing in action. Ambiguous loss creates a general sense of confusion which hinders the ability to grieve the loss of the family member (Boss, 1999).

Boundary ambiguity is a contributing factor of ambiguous loss. Who is in or out of the family is determined by family roles and rules. Increased boundary ambiguity can lead to a rise in negative outcomes such as depression and anxiety. Families that are able to reframe individual member roles and create meaning are able to decrease boundary ambiguity (Berge & Holm, 2007). A lack of closure keeps families from seeking new meaning, developing coping skills, and ultimately adapting to change (Boss, 2007).

THEORY SUMMMARY

The use of three theories is necessary to analyze how each caregiver deals with her child's diagnosis and its ramification on the overall family life. The FAAR Model highlights the resources and stressors that each caregiver identifies during her interview. The Model also shows how different caregivers adjust and adapt to sickle cell disease. Life Course Theory helps to identify differences between the caregivers and their children who are at different ages and life stages. The burden of care may be different for the caregivers as their children meet new life stages. The concept of time also is of high importance to caregivers as they reflect on pre Sickle Cell diagnosis and post Sickle Cell diagnosis. Finally, Ambiguous Loss Theory will shed light on any grief the caregiver displays and the effects of loss on family life. Family loss also may be

amplified or decreased in conjunction with caregiver or child life stage and the resources available for adaptation.

FIGURE 1
MODEL OF SICKLE CELL DISEASE AND CHRONIC SORROW

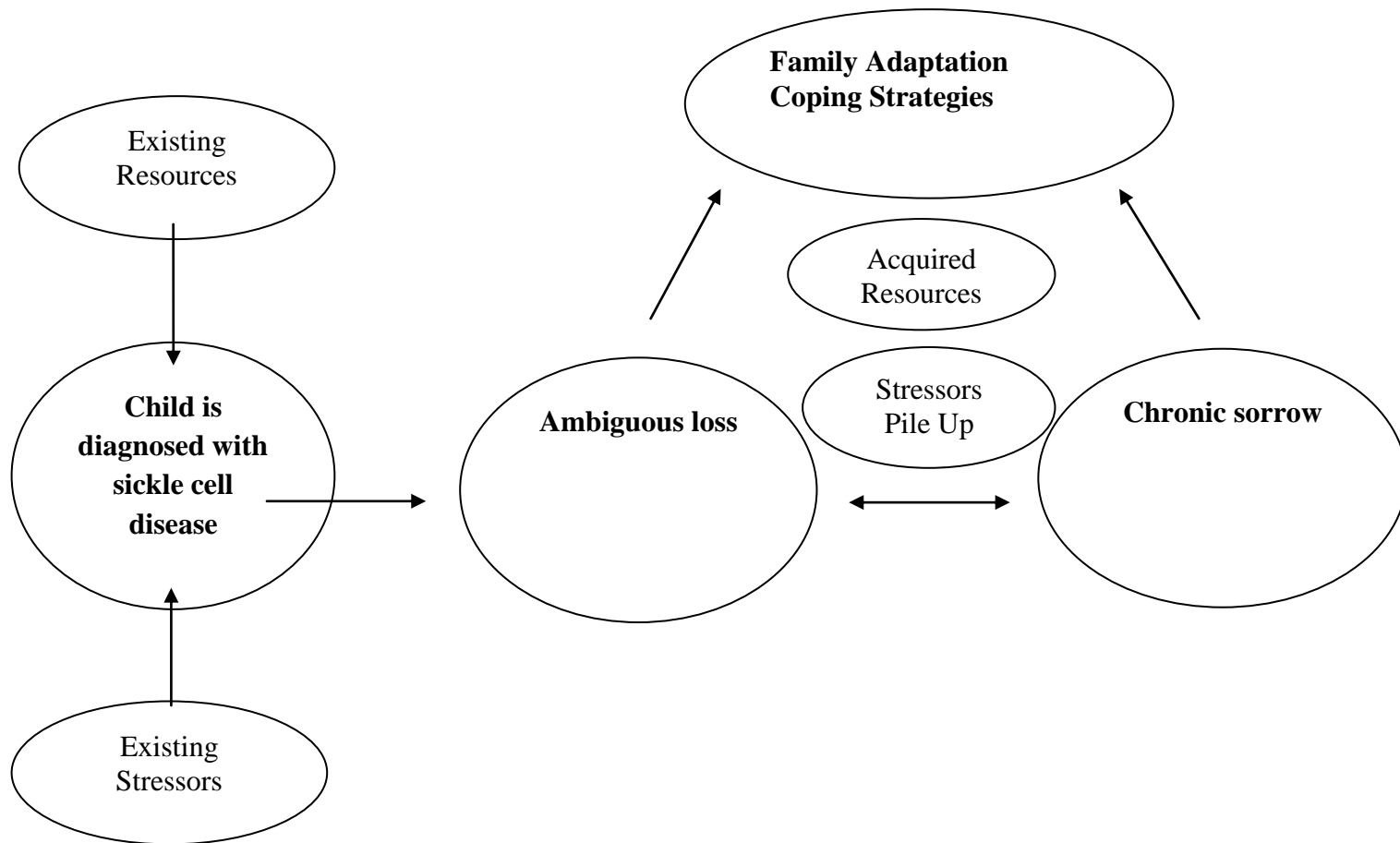


TABLE 1**INTERVIEW QUESTIONS IN RELATION TO RESEARCH QUESTIONS**

The following table illustrates the purpose of the interview questions in relation to the previously identified research questions.

| | How is chronic sorrow a factor in daily family functioning? | How do caregivers cope with the unknown future of their child's sickle cell disease? | What are experiences with the medical team? |
|--|---|--|---|
| Tell me about yourself and your family. | X | | |
| Tell me about (blank), your child with sickle cell disease. | X | | |
| When did you first learn about your child's diagnosis? | X | | |
| How did you and your family react to the news about sickle cell disease? | X | | |
| How did you feel after receiving the new diagnosis? | X | | |
| What were your biggest worries when your child was first diagnosed? | X | X | |
| In what ways did your family adapt? | X | X | |
| What does a good day look like at your house? | X | X | |
| What does a bad day look like at your house? | X | X | |
| Do you have sources of support that have helped you? | X | X | |
| Have these sources of support changed over the years | X | X | |
| What have been your challenges? | X | X | |
| How did you respond to these challenges? | X | X | |

TABLE 1 (CONT'D)

| | How is chronic sorrow a factor in daily family functioning? | How do caregivers cope with the unknown future of their child's sickle cell disease? | What are experiences with the medical team? |
|--|---|--|---|
| How did you respond immediately? | X | X | |
| How did you respond over time? | X | X | |
| Has your child been hospitalized before? | X | X | X |
| Did hospitalization create new challenges? | X | X | X |
| Did your feelings resurface or change after experiencing new challenges? | X | X | |
| What helped you manage your emotions? | X | X | |
| What have been your feelings about the medical team? | | | X |
| What was helpful? | | | X |
| What was not helpful? | | | X |
| What does sickle cell disease mean to you today? | | X | |
| What are your biggest worries today? | | X | |
| What do you hope for your child and family in the future? | | X | |

CHAPTER TWO

REVIEW OF THE LITERATURE

Chronic sorrow has been studied in caregivers across a variety of acute and chronic illnesses, yet there continues to be lack of replicated data on specific populations. A groundbreaking study by the Nursing Consortium for Research on Chronic Sorrow (NCRCS) found that 82% of family caregivers exhibited signs of chronic sorrow in response to having a family member with a chronic illness (Burke, Eakes, & Hainsworth, 1999). To date, there is only one published study regarding the feelings of chronic sorrow in African American parents of children with sickle cell disease (Northington, 2000).

CHRONIC SORROW

As cited by Roos (2002), the concept of chronic sorrow was first described in 1962 by Olshansky. Olshansky noted the emotional struggles of parents raising children with “mental retardation.” Chronic sorrow represents the paradox of feelings surrounding the reality of the illness and the dreams for a normative childhood. The wishes and dreams of childhood are constantly being revised by illness demands, and parents must continually put forth energy to adapt the family system.

DEVELOPMENTAL MILESTONES AND NEW SYMPTOMS

Missed developmental milestones can be a trigger for parental episodes of chronic sorrow. Parents may regress to their initial stages of grief during diagnosis and again grapple with the fantasy of what life could have been. Normative childhood development may not be possible because of the effects of chronic illness. Developmental triggers may be predictable and include missing the stages of walking, talking, going to school, entering puberty, and leaving the home during young adulthood. With more complex diseases, parents also may have to grapple

with out of home placement and guardianship concerns in case of parental death. Siblings or other family members surpassing developmental milestones of the chronically ill child also may trigger an episode of chronic sorrow (Roos, 2002).

In a study of parents of children with cystic fibrosis, over fifty percent of parents felt they lacked information regarding their child's developmental needs. Developmental information from the medical team can help parents to understand how illness affects development and how tasks can be adapted for each child (Hymovich & Baker, 1985). Anticipatory guidance surrounding developmental milestones and chronic sorrow can greatly empower parents to maintain family equilibrium or make small changes towards system adaptation. The feelings of stress and loss during acute crisis may not be predictable, but parents can make small changes and rally resources to accommodate the stress of missed developmental milestones. Milestones that trigger episodes of loss are not always detrimental, but can be a time for great family strength and resilience (Roos, 2002).

New symptoms also may increase the likelihood of an episode of chronic sorrow because they may indicate disease progression or may highlight the uncertainty surrounding the future of the chronic illness. Parents may feel lack of mastery and may question their ability to provide appropriate care for the child. Parent concerns that are dismissed by the medical team can decrease family mastery and hinder family adaptation to illness (Bettle & Latimer, 2009).

CHRONIC ILLNESS AND THE FAMILY

Many parents strive for normalization through the course of illness. During periods of optimal health and family stability, medical care may seem minimally disruptive. Family routines take priority over medical routines. Family mastery over illness is increased and feelings of control may return to the family (Bluebond-Langer, 1996). Normalization also helps

to remind parents of what the ill child can do, not what they cannot do. The ill child may be treated the same as well children in the family. Rules regarding discipline can be instituted during times of optimal health. Parents noted uncertainty regarding whether misbehavior was because of the illness or a normal developmental response. Discipline helps to remind the ill child of limits on behavior which can then be reinforced during times of crisis (Canam, 1993).

Children affected by chronic illness also may have good days during which they appear to be functioning optimally. This creates confusion for other family members regarding the level of stress and ambiguity that illness creates within the family. This also can create false illusions of mastery for the family system. Outside support systems and resources may not be fully utilized because of the false appearances of the child and family coping well (O'Brien, 2007).

As an illness progresses, it becomes harder for parents to normalize the situation. An episode of chronic sorrow may be triggered by the futility of maintaining normalization for the family. Sudden lack of mastery and control surrounding illness progression finds many parents regressed in their overall coping abilities (Bluebond-Langer, 1996). Periodic episodes of chronic sorrow may be an indication that parents are again cycling through resistance, restructuring, and adaptation. As an illness progresses, new demands are placed on the family and change must occur again. Parents also may be searching for new meaning within their child's illness as the disease progresses or the child misses more developmental milestones (Barnett, Clements, Kaplan-Estrin, & Fialka, 2003). The Nursing Consortium for Research on Chronic Sorrow conducted two studies involving caregivers of individuals with mental illness or multiple sclerosis. Overall, the burden of continual care was a key trigger because it often highlighted the loss of family norms. Many families also were able to note specific events or anniversaries that

triggered feelings of chronic sorrow. Some of these anniversaries triggered the feelings of devastation surrounding the person's initial diagnosis (Burke, Eakes, & Hainsworth, 1999).

Cousino and Hazen (2013) conducted a systematic review of the literature on parenting stress. They noted that caregivers across a variety of chronic illnesses noted greater parenting stress than parents of healthy children. The burden of continually caring for an ill child creates stress across the lifespan. Parents who perceived their ill child as increasingly vulnerable showed higher levels of parenting stress. Parents also were more likely to bring their child in for more frequent and often unnecessary medical exams due to the feelings of vulnerability for their child (Cousino & Hazen, 2013).

GENDER DIFFERENCES IN CAREGIVERS

Research by Mallow and Betchel (1999) found gender differences in the periodic episodes of chronic sorrow felt by parents of children with developmental disabilities. Mothers were more likely to experience episodes of sadness and grief during healthcare crises. These times of medical complication and possible illness progression evoked episodes of sorrow and renewed the uncertainty surrounding the child's future. Mothers are often the main caregivers for the ill child, so a change in prognosis also can create role confusion (Mallow & Bechtel, 1999). Mothers also were more likely to self blame and internalize feelings of chronic sorrow (Roos, 2002). In contrast, fathers tended to experience sadness as the child missed normative developmental milestones. Fathers who are traditionally breadwinners in the family are responsible for planning and providing for the future. A child with chronic illness may add an element of uncertainty to the future of the family system. Children with a chronic illness also may add financial stress that fathers may have to manage (Mallow & Bechtel, 1999). The long term stress surrounding the future of a child's chronic illness can cause periods of chronic sorrow

for family members and freeze their ability to grieve new losses associated with the illness. (Boss, 2007). In addition to gender differences, feelings of loss and the intensity of chronic sorrow often differs between family members. This can be a source of tension if each member is not able to comprehend individual stressors and coping strategies. Each family member must reconstruct a new identity in the face of illness and the cohesion of these identities shapes the family as a whole (Roos, 2002).

Mothers of children with sickle cell disease were more likely to become overwhelmed with their child's care than fathers, and intense engagement often occurred when the physical burden of illness was high. Times of low illness burden often reminded mothers of the sorrow associated with a child with disability. Mothers reflected on the uncertainty of the child's future when the child was well and having few complications from sickle cell disease (Atkin & Ahmad, 2000). Overwhelming feelings of loss also occurred when social support and resources were low. Mothers reported feeling overwhelmed as a normative reaction to the daily stressors surrounding illness. Coping with sickle cell disease led mothers to contemplate the untimely death of their child, separation anxiety from the ill child and well family members, and social isolation from the general population (Burnes, Antle, Williams, & Cook, 2008).

CHRONIC ILLNESS AND AMBIGUOUS LOSS

The uncertainty of chronic illness creates a situation of ambiguous loss for parents. Despite medical advances, children with sickle cell disease still have an unclear prognosis and may have a limited life expectancy. Triggers for chronic sorrow can be internal or external, and episodes of sorrow may intensify as a child's illness progresses (Burke, Eakes, & Hainsworth, 1999). In one study, parents of children with sickle cell disease noted that internal triggers of chronic sorrow often related to dreams for their child's future. Parental reflections on the

limitations of sickle cell disease led parents to renew previous feelings of sorrow similar to the time of diagnosis. Parents also noted that external triggers of chronic sorrow often were related to illness complications and health restrictions (Northington, 2000).

Ongoing loss associated with chronic illness disrupts the parent child relationship. The intensity and attachment between parent and child is affected by the illness and how it affects daily life. Parents experience this grief so profoundly because of the hopes and dreams they had for a normative childhood (Teel, 1991). Parents feel not only loss of the perfect child, but also self loss of the life they thought they would have. Self loss and other loss can compound to intensify feelings of chronic sorrow and can greatly hinder family adaptation (Roos, 2002). Parents who become engulfed with the caring of their child may struggle with lack of support and resources. The ability to set boundaries between the roles of parent and caregiver can help parents to maintain a sense of self (Atkin & Ahmad, 2000).

Increased boundary ambiguity is correlated with increased parental distress and mental health issues. Boundary ambiguity is a factor of ambiguous loss that can be increased or decreased based upon family adaptation (Berge & Holm, 2007). Research by Mu and Tomlinson (1997) focused on family crisis during acute illness and sheds light on the uncertainty of chronic illness and the boundary ambiguity that occurs during initial diagnosis. Mu and Tomlinson (1997) noted that this initial boundary ambiguity and the future implications of the illness forced parents to reevaluate family functions and demanded family restructuring. The longer parents remained uncertain about the diagnosis and prognosis, the longer the initial ambiguity lasted and therefore increased parent role strain. Thus, chronic illnesses with clear management courses and minimal daily disruptions decreased uncertainty and initial ambiguity. Mu and Tomlinson (1997) also propose that family systems that learn to self regulate during crisis to manage the ambiguity

return to family equilibrium more quickly. Therefore, normalizing family life and maintaining interpersonal relationships helps to maintain the family system's integrity (Mu & Tomlinson, 1997).

EXPERIENCES WITH THE MEDICAL TEAM

Tomlinson and Harbaugh (2004) concluded that uncertainty regarding illness prognosis, lack of information from the medical team, and change in function of roles increases boundary ambiguity. Consequently, acute hospital admissions to manage chronic illness can dramatically increase boundary ambiguity and increase episodes of chronic sorrow. New medical caretakers may question roles and decision making abilities that previously were functioning and maintaining equilibrium in the family (Tomlinson & Harbaugh, 2004). Parents who were unable to get accurate information regarding their child's care often felt helpless and lacked confidence in their caretaking abilities. This lack of confidence affected overall family adaptation. Parents who developed a positive relationship with the medical team and learned how to navigate the healthcare system increased family coping abilities (Canam, 1993).

Feelings associated with ambiguous loss may worsen when children spend lengthy periods of time in the hospital or in other long term care. Children born with complex chronic illnesses may spend lengthy periods of time in the hospital after birth, and they may be psychologically present but physically absent from the family. Prolonged hospitalization can interrupt parent-child attachment as well as delay effective coping for the family. The family system will likely face increased stress surrounding the ambiguity of illness until the child comes home and adaptation processes begin (Roper & Jackson, 2007).

Acute hospitalizations also are a reminder to parents of the progression and complications associated with illness. Illness crisis may send parents into an episode of chronic sorrow as they

come face to face with other children with more advanced disease progressions. The medical team also may question home medical routines and explore concerns of lack of adherence. The questioning of parent roles often leads to increased role strain and boundary ambiguity (Bluebond-Langer, 1996). Parents of children with sickle cell disease noted the lack of support from the medical team and the incompetence surrounding hospital admissions. The lack of trust between the parents and healthcare provider increased family stress and sorrow. Parents also noted the lack of information surrounding their child's disease (Atkin & Ahmad, 2000).

A study about family communication and the adolescent with sickle cell noted the lack of knowledge regarding sickle cell disease and healthcare workers. Parents felt a need to advocate for their adolescent child because of the varying levels of incompetence and lack of knowledge at different hospitals. This increased feelings of mistrust towards healthcare workers. To minimize feelings of distrust, parents often tried to seek out a hospital that was familiar with sickle cell disease or familiar with their child (Graff, et al., 2012).

Distrust between the family and the medical team also can occur when there is disagreement surrounding pain assessment. This disagreement is often a lack of understanding of how each child and family reacts to sickle cell disease. Parents who lacked a trusting relationship with the medical team were more likely to feel their child was incorrectly assessed for pain. Parents reported a lack of urgency for their child's care and were questioned regarding their child's drug addiction behaviors. Children may learn to overestimate their pain scores if they are fearful of pain medication being withheld. Parents and children can form an alliance with the medical team and learn to accurately assess pain and have their needs met during a medical crisis. Parents who are able to communicate previous positive and negative experiences regarding their child in pain may help the medical team provide better care (Beyer, 1999).

Hospitalizations where the parents perceived the child as being increasingly vulnerable and ill were more likely to increase boundary ambiguity than less serious hospitalizations. This point highlights Boss's findings that a family member's status as in or out of the family system increases distress and ambiguity (Tomlinson & Harbaugh, 2004).

Roos (2002) suggests that healthcare teams that focus on partnership with families are better able to promote self efficacy and empower the family to continue coping and adapting. A strength based approach to physician-family collaboration increases feelings of trust and mastery. Medical teams that force parents towards acceptance and closure are likely to note the negative aspects of family regression instead of the positive aspects of adaptation. Chronic sorrow can be manageable for parents when they feel a sense of control over the illness and respect from the medical team (Roos, 2002). A key element of parental control was the ability to question medical authority. Parents felt more confident to question medication and treatment changes because of their expertise as caregivers at home. Some parents also noted that increased self confidence diminished the belief that doctors are infallible. Parents who were empowered through self confidence found deeper meaning and in turn felt they had more control in their child's care (Jerrett, 1994).

Healthcare teams that provide parents with anticipatory guidance surrounding disease progression are optimizing long term coping abilities for the family. Some chronic illnesses include a negative progression of symptoms and increased medical management. New developmental stages for the child and illness progression may have greater implications for the family system. Anticipatory guidance may help to decrease the pile up of stressors that trigger prolonged episodes of chronic sorrow. Helping parents to brainstorm coping strategies and to

deal with new stressors of the illness will help families continue to adapt and manage the ambiguous loss associated with illness (Ahmann & Rollins, 2005).

COPING STRATEGIES

The ambiguity of chronic illness can make it difficult for parents to cope effectively and function well as a family (Berge & Holm, 2007). Meaning making is an essential part of family coping and adaptation. During episodes of chronic sorrow, parents may have to search for new meaning, and find hope within the unknown. Each family member must find meaning with the chronic illness in order for the whole family system to move forward (Boss, 1999). Meaning making is a necessary component to managing the emotional reaction of caring for a chronically ill child. Meaning making helps parents to develop new coping styles and process illness related changes (Jerrett, 1994).

Some parents of children with sickle cell disease noted a change in perspective that allowed them to balance the ambiguity of illness. These parents learned to continually adjust their attitudes and adapt family functioning when needed. This new perspective helped some parents to avoid prolonged episodes of chronic sorrow (Northington, 2000).

Prolonged episodes of chronic sorrow may decrease a parent's overall coping abilities and long term resources for family adaptation. Parents who ineffectively cope with the burden of illness also may increase stress for the ill child. Long term experiences of illness related stress can lead to a decrease in the child's self concept and an increase in behavior issues. Prolonged episodes of chronic sorrow also may lead to mismanagement of the child's healthcare (Lutz, Barakat, Whitley, & Frempong, 2004).

Parental locus of control has been shown to affect the child's quality of life. Parents who note an internal locus of control while managing their child's sickle cell disease were more likely

to encourage higher levels of self competence and self esteem for their child. However, the majority of primary caregivers of children with sickle cell disease noted an external locus of control. Frequent need for medical intervention and lengthy inpatient hospitalizations for pediatric patients may take control away from caregivers. An external locus of control can decrease overall family functioning and interfere with family adaptation (Barakat, Lutz, Nicolaou, & Lash, 2005).

A study of parental stress found that parents with poor communication and lack of support felt increasingly vulnerable and stressed. Resources and other resistance factors that assist in adaptation were lacking because of poor coping strategies. Parents who practiced open communication and positively adjusted to their child's illness used problem solving coping strategies. The problem solving coping strategy was increasingly helpful during changes in the child's health status (Hall, Neely-Barnes, Graff, Krcek, & Roberts, 2012).

Parents who adopted "day to day" approaches to coping and providing care were less likely to become engulfed in the sorrow of having a child with sickle cell disease. Those with this approach were able to normalize the situation and indicated positive aspects of the disease for their family (Atkin & Ahmad, 2000). The coping strategy of living in the present can have negative ramifications when parents refuse to process new medical information or are unwilling to plan for future medical complications (Dodgson, Garwick, Blozis, Patterson, Bennett, & Blum, 2000). Many parents also continue to use coping strategies that were effective in the early stages of diagnosis, and those who are unable to adopt new coping techniques for future crises may face larger devastation when initial strategies become ineffective (Langridge, 2002). Feelings of chronic sorrow can be exacerbated when the fantasy of the future takes precedent over the reality of the illness (Roos, 2002).

Action strategies for coping with chronic sorrow have been proposed to increase parental sense of control. For example, role and identity confusion created by chronic illness can be minimized when parents are able to maintain a sense of self through hobbies. Parents who continue to devote personal time to hobbies and interests may cope more successfully when caregiver stressors arise. Parents who maintain their personal mental health and seek respite are better able to help the overall family cope and adapt with illness (Lee, Strauss, Wittman, Jackson, & Carstens, 2001).

Boss (2007) proposes that families adopt both/and thinking to help manage the stress of ambiguous loss. Both/and thinking can help promote family resiliency during times of crisis. Parents who are able to comprehend that their child may have a shortened lifespan due to illness, but still have hope for future medical advances and cures are better able to manage the ambiguity that illness creates. Healthcare teams should never extinguish a parent's hope for the child's future, even when positive outcomes seem impossible (Boss, 2007). Helping parents maintain hope discourages self blame and hopelessness that can intensify feelings of chronic sorrow and ambiguous loss. Parents who hope for closure or an end to the repercussions of illness are likely to become stagnant in the adaptation process. Hope can bring positive or negative growth to a family, dependent on their ability to reconstruct a new family identity and temper their sense of mastery for the future (Boss, 2006). Parents who maintain a sense of hope are likely to also instill a sense of hope in their ill child. Children who maintain a sense of hope may also encourage their parents during times of crisis (Venning, Eliot, Whitford, & Honnor, 2007).

Both/and thinking also helps parents vacillate between viewpoints during times of optimal and poor health. Parents who are able to experience periods of illness mastery and adaptation are better able to balance multiple viewpoints of their illness during crisis. Parents

may shift their feelings of hope for a cure to hope for better illness management (Bluebond-Langer, 1996). Families that are able to identify previous coping strategies may find insight to their inner strengths and find new resolve in the functioning of their family. A sense of control and mastery over previous crises helps parents to develop the ability for mastery of future crises. Complete mastery is never possible because of the ambiguity of illness, but helping parents to learn to temper their sense of mastery instills feelings of control and boosts success for family adaptation (Boss, 2006). Anticipatory guidance is a more effective means of supporting family adaptation than forced adaptation during major crisis (Ahmann & Rollins, 2005).

Mastery of a chronic illness is almost impossible because of the ambiguity. Parents that reframe the situation and accept that final outcomes are unknown are able to reduce their anxiety and decrease episodes of chronic sorrow. Meaning making is an essential part of family coping and adaptation. During episodes of chronic sorrow, parents may have to search for new meaning, and find hope within the unknown. Each family member must find meaning with the chronic illness in order for the whole family system to move forward. Both/and thinking can be an effective tool to promoting overall family resiliency and adaptation (Boss, 1999).

FAAR MODEL

A parent's ability to cope well and manage feelings of chronic sorrow is largely dependent upon the family's ability to adjust and adapt to the child's illness. The demands and stressors of a child with a chronic illness are high and can be intensified with hospitalizations and medical complications (Patterson, Holm, & Gurney, 2004). The cumulative effect of illness related family and daily life stressors make family coping and adaptation seem impossible. Family resiliency decreases as the pile up of stressors overwhelms the family system (Boss, 2006). Previous studies of chronic sorrow have indicated parental feelings of lack of resources

and support from the healthcare team. Interventions aimed at providing support and enhancing coping skills would be beneficial if triggers of chronic sorrow for parents of children with sickle cell disease could be identified (Northington, 2000).

Upon initial diagnosis, the family may resist making changes to their family functioning. This resistance is often accompanied by denial of the illness, and questioning of the child's future. Most families begin to restructure their family functioning and make small changes to the family system because of the numerous demands the illness has on the family as a whole. Parent roles must change to accommodate daily medical care and illness awareness (Patterson, Holm, & Gurney, 2004). Parents dealing with rare or undiagnosed illnesses may feel social stigma and lack of support. An illness that cannot be identified also may be a loss that is unidentifiable. This lack of support from society feeds into feelings of chronic sorrow and intensifies ambiguous loss (Roos, 2002).

Families who are already vulnerable because of the stressors of illness are likely to feel frustration in response to other life stressors. Relationship maintenance and work commitments may seem impossible to manage when enmeshed within the effects of illness. The pile up of stressors also may highlight the compounding effect of other losses (Roos, 2002).

The final stage of the FAAR Model is adaptation. Parents must adapt their family functioning and parenting skills to meet the needs of their child and medical condition (McCubbin & Patterson, 1983). The ability to find deeper meaning with the illness is an important step towards family adaptation. Meaning making facilitates coping and helps families move forward with their child's illness (Mu & Tomlinson, 1997). Adaptation does not occur smoothly for all families and is largely dependent upon support systems and resources already in place. Families with limited resources are likely to struggle with adaptation to the illness

(McCubbin & Patterson, 1983). An estimated one third of families are unable to find a balance between the stressors and resources available. These families may struggle with increased interpersonal conflict and new mental health issues. Parents may have to continually manage stressors and change family functions in response to continual need for adaptation as an illness progresses (Barnett, Clements, Kaplan-Estrin, & Fialka, 2003).

CHAPTER THREE

METHODOLOGY

This case study analysis examined the primary caregiver of two children ages three to twenty one years with sickle cell disease who receive medical care at Hurley Medical Center Sickle Cell Clinic. This investigator conducted two semi-structured interviews of African American primary caregivers of children treated at Hurley Medical Center.

The caregiver interviews represent a combination of two case study methods. Intrinsic case study design allowed the primary investigator to learn more about the generalities of being a caregiver. The primary investigator's desire to learn more developed the thesis topic. The purpose of the study was not to develop theory, but to learn about caregiver struggles and successes. The caregiver interviews also could be characterized as an interpretive case study. The goal of this method was for caregivers to tell their story with sickle cell disease. Open ended interview questions encouraged caregivers to lead the direction of the story. Giving voice to the caregiver was meant to be empowering. The interpretive case study method amplified the details shared which provided more data for constant comparison analysis (Stacke, 2000).

The case study approach provided rich contrasting narrative of caregiver experiences as they cared for their child with sickle cell disease. With an understanding derived from parental care narrative, medical staff can be better informed to increase family and child supports during times of optimal health and during crisis. Parent empowerment through case study narrative also helped to highlight the importance of resources and supports for the family. Acknowledgement of family resources and supports was important to draw the family together during crisis and adapt to disease related changes more efficiently.

SAMPLE

Hurley Medical Center is located within the city of Flint, Michigan, but also provides medical care to children residing in greater Genesee County. In 2007, the median income for the city of Flint was \$26,143, in contrast to the median income of Genesee County which was \$43,112. U.S. 2008 Census data for Genesee County reported 16.6% of the population was living below the poverty line (State and County Quickfacts, 2010). Results of the 2007 American Community Survey found that 35.5% of the city of Flint lives in poverty (Bishaw & Semega, 2008). Low median incomes and high poverty rates may increase stressors for caregivers of children with sickle cell disease. Resources are often limited for those with income restrictions and can correlate with poor health management. Currently, there are 113 children with sickle cell disease who receive treatment at Hurley Medical Center and four children who receive monthly exchange transfusions.

Primary caregivers of children who receive monthly exchange transfusions were identified by Sickle Cell Clinic staff. Children with an increased risk of stroke or past history of stroke receive an exchange transfusion every four weeks at Hurley Medical Center Sickle Cell Clinic. These children are typically older and years past their initial diagnosis of sickle cell disease. The goal of three cases was set with the hopes of interviewing parents of children in early, middle, and late childhood. A total of three cases were to be selected within ninety days after distributing the invitation letter. When this researcher was unable to obtain three cases from Sickle Cell Clinic, a caregiver of a child with sickle cell disease from a general hospital admission was interviewed. Sickle Cell Clinic staff provided primary caregivers with an informational letter introducing the need for continued understanding of how primary caregivers care for their child's sickle cell disease. The letter indicated research collaboration between the

Hurley Child Life Department, Hurley Pediatric Hematology Department, and Michigan State University. The investigator invited caregivers to participate in the research study and encouraged them to contact the investigator to tell their story about sickle cell disease.

IRB PROCEDURES

IRB approval was obtained from both Michigan State University and Hurley Medical Center. Michigan State University IRB classified the study for exempt review. Hurley Medical Center classified the study for full review. This investigator was required to present the study before the Hurley Medical Center IRB board before gaining approval. Both institutions requested revisions to the methodology. A secondary approval was requested of both IRB groups to extend the recruitment period another 120 days due to lack of participation. Several delays were encountered due to differences in the institutions scheduled IRB meeting dates. While revisions were made in a timely manner, a month waiting period was often needed before gaining approval on revisions at each institution. This investigator also took an extended leave of absence from work due to a family emergency. During this two month leave of absence, no recruiting was possible and potential cases were missed. This leave of absence was unavoidable but did have an effect on the ability to recruit and decreased continued interest in the project from Hurley Medical Center staff.

RECRUITMENT

Eight informational letters were handed out in Sickle Cell Clinic by staff during the initial ninety day recruitment period. One caregiver from Sickle Cell Clinic agreed to participate. A Sickle Cell Clinic staff member notified the investigator of the caregiver's willingness to interview. The first interview was completed in the Sickle Cell clinic during the patient's scheduled exchange transfusion appointment. The caregiver initially forgot about her interview

appointment. This interview was rescheduled for a month later during the patient's next exchange transfusion appointment.

During the second recruitment period, a caregiver was recruited from Hurley Medical Center Pediatric Unit. The caregiver was identified by staff on the general pediatric unit and was willing to complete the interview within the hour of being approached. Two additional informational letters were distributed to caregivers on the general pediatric unit, but no further interviews were completed. Therefore, only two caregiver interviews were completed for this study.

Caregivers were instructed to contact the primary investigator if they wished to participate in the study. Caregiver one was recruited through Sickle Cell Clinic staff. Caregiver two was recruited through Pediatric Unit staff. Caregivers were informed of study risks and limitations. Caregivers may be familiar with medical consent forms due to the nature of their child's medical needs. The investigator assisted caregivers with reading and comprehension of the research consent form as necessary. After obtaining informed consent, interviews with primary caregivers were conducted at Hurley Medical Center Sickle Cell Clinic during an outpatient exchange transfusion appointment or during the child's hospital admission.

INTERVIEW CONTEXT

The interview questions were developed using the *Burke/NCRCs Chronic Sorrow Questionnaire* (Burke, Hainsworth, Eakes, & Lindgren, 1992). The questions explored the child's diagnosis, family challenges, family supports, and the experiences with the medical team. The interview questions were intended to shed light on the research questions identified in chapter one. Please reference Appendix D: Caregiver interview.

The first interview was conducted by the primary investigator in a private conference room at Hurley Medical Center Sickle Cell Clinic. There were no additional members of the research team or any staff members of Hurley Medical Center in the conference room during the interview. The conference room had a large table and chairs which created distance between the caregiver and the investigator. While the conference room provided more privacy, it also added a level of formality. The conference room was located adjacent to the patient's clinic room which allowed the caregiver to take frequent breaks to check on her child's treatment. The interview on the general pediatric unit was conducted in the Child Life office. This office is located adjacent to the playroom and main entrance which created additional noise and disruption. No additional staff members were in the office during the time of the interview. A note was placed on the door to minimize interruptions. The Child Life office has no table, but instead two office chairs were grouped across from one another to facilitate conversation. The office also was located within close proximity of the patient's room, but the caregiver requested no breaks and did not appear concerned about the distance from her child.

DATA COLLECTION

The interviews lasted no longer than ninety minutes and were semi-structured to better allow caregivers to explore feelings surrounding sickle cell disease. Themes were drawn from the narrative to evaluate feelings associated with care for a child with sickle cell disease. The *Burke/NCRCs Chronic Sorrow Questionnaire* was used as a reference point to develop interview questions, but was expanded to encourage exploration of feelings regarding the effects of sickle cell disease on the family. The *Burke/ NCRCs Chronic Sorrow Questionnaire* was devised to investigate themes of burden of care, triggers of chronic sorrow, and interventions aimed at improving coping skills. The validity and reliability of this questionnaire was determined in the

original study of parents with children with myelomeningocele by Mary Burke in 1989 (Burke, Hainsworth, Eakes, & Lindgren, 1992). Additional interview questions were presented to encourage caregivers to share their story. Parental responses from the semi-structured interview were audio recorded, transcribed, and analyzed.

DATA ANALYSIS

Both cases were transcribed by a professional transcriptionist who lived outside of the recruitment area. This was done to maintain privacy and decrease chances of error. The primary investigator edited the transcription again to correct physician names and local idioms unbeknown to the transcriptionist.

The constant comparison method was used in data analysis. The constant comparison method was developed by Glaser and Strauss in 1964. This qualitative method was chosen because it allows the investigator to code for themes present in the transcription material and create a theory that may explain the data. This investigator was not looking for the causality of chronic sorrow, but instead wished to explore themes within the topic. The constant comparison method does not generate universality to apply to all phenomena in similar topics (Glaser, 1965).

There are four stages of the constant comparison method. In the first stage, the material is coded into categories. Every new piece of information in each category is compared and placed in the same or a new category. This initial phase helps the investigator to reflect on all aspects of the category and how each piece of information fits or does not fit the category. The second stage of analysis integrates categories and compares incidents within the same category. Within each specific category, each incident is compared and contrasted. This allows the researcher to better understand the diverse properties of each category. It may also shed light as to how categories are related and a theory may develop. The third phase delimits the theory. As the theory develops

and solidifies, there are fewer incidents that will change the overall development of the category. As it becomes clear how categories are related, the investigator will be able to narrow down the original list of categories. A clearer boundary of the theory allows the investigator to eliminate data that does not apply or has already been coded. The fourth phase of constant comparison invites the researcher to write the theory that the data created. The themes of the theory are pulled from the coded categories (Glaser, 1965).

Both interviews were coded using the constant comparison method. Themes were noted in the interview transcription and put into categories. Content analysis for the case study narrative was coded to recognize emerging themes such as those from FAAR and ambiguous loss literature. Narrative was coded for themes surrounding family adaptation, emotions surrounding illness, and coping skills. Cross case analysis and discussion was completed to highlight any themes that may emerge between individual cases.

The primary investigator sought to address trustworthiness of the data by using interview questions that had been previously tested in a groundbreaking study by Mary Burke (Burke, Hainsworth, Eakes, & Lindgren, 1992). The primary investigator also evaluated her credibility and reflexivity as a researcher. My background as a Certified Child Life Specialist has led to extensive training in speaking with parents. This training encourages supportive listening to better elicit parent responses. The contrasting data suggests the applicability and transferability to other caregivers of children with sickle cell disease. It is likely that other caregivers have successfully adapted to sickle cell disease and that other caregivers still struggle with the continual changes (Krefting, 1991).

ROLE OF THE RESEARCHER

The idea for this thesis topic came through my experience five years ago while working on the east coast in a large metropolitan teaching hospital. As a Child Life Specialist, I was part of the treatment team taking care of a two year old female who had a degenerative muscular disease. While cognitively appropriate for her age, her body was unable to complete simple movements. As her Child Life Specialist, I spent many hours with the patient providing her with emotional support when having a bad day or activities that could match her physical abilities. I began creating legacy projects as the child's illness progressed.

The patient's mother lived almost two hours from the hospital and was frequently away because of the need to balance work and hospitalization. The mother's grief for her child often occurred during new admissions or changes in the treatment plan. Her positive attitude would slip away to defiance and anger. The turning point for this patient came when she had spent over 250 of the 365 days of the year at the hospital. The treatment team decided that her illness had progressed to the next step and a tracheotomy was required for airway support. The mother's grief was so raw she was unable to make a decision for her child. For another month, no progress was made towards a tracheotomy, and the mother avoided the hospital at all costs. A decision to move forward was ultimately made when the patient's mother was ready for the changes ahead. She began completing insurance paper work, was trained in respiratory care, and moved homes so that her child could have better home nursing support.

I have never forgotten this patient and her mother. I now see coping with chronic illness through a lens that requires continued change and adaptation. My current role as a Child Life Specialist in a Midwest children's hospital provides me with the rare opportunity to help children and their families understand and cope with the hospital experience. I have had numerous

conversations with parents regarding the emotional burden that comes with caring for a child with a chronic illness. I also have witnessed how change in medical care can suddenly seem impossible to the once confident caregiver.

This qualitative research will give the medical team better insight into how the family adapts to illness. It also will also give voice to parents and empower them to continue to provide care for the child with sickle cell disease and interface with the medical team. My psychosocial role on the medical team calls for me to improve my clinical skills through evidence based research. Parents who are burdened by feelings of chronic sorrow may benefit from additional support from an informed medical team. Additional resources may be available for families who are struggling with adaptation. To truly provide children with family centered care, we need continued research and support for their caregivers.

CHAPTER FOUR

RESEARCH FINDINGS

CAREGIVER ONE: PAMELA

Pamela is a forty six year old married woman. She is a college graduate and is currently employed as a logistics manager. Pamela and her husband have an annual household income above \$65,000. Pamela has three children ages nineteen, seventeen, and eight years old. Her nineteen year old son Ryan is the only child with sickle cell disease in the family. Ryan currently attends a state university several hours from home and lives independently on campus. This interview was conducted on the Hurley Medical Center Pediatric Unit where Ryan was admitted for medical care. Pamela was approached for the interview at the patient's bedside.

THEMES

KNOWLEDGE OF DISEASE

Pamela noted that Ryan's diagnosis with sickle cell disease was far from ordinary and largely uneventful. Ryan was not diagnosed at birth like most children but instead at age eight when he needed routine blood work for an upcoming surgery. She stated:

So his fetal hemoglobin was so high that it masked the disease so it didn't present itself ... he had it since he was born but it didn't present itself until he was like 8. So, for 8 years it was just business as usual – just raising a little boy.

Pamela stated that Ryan had his first sickle cell crisis at age ten and has been hospitalized about once every year. She credits her understanding of sickle cell disease to the two medical providers that have taken care of Ryan his entire life. She noted:

I mean they're that familiar with him and they've been just wonderful to him throughout this whole process. Explaining things, getting us through different aspects of the illness...a lot of the hospitalization...talking to us..being able to explain things in a way we can understand it and what's going on. But they've put our mind at ease about a lot of things and they've helped him through a lot of quiriness that has come with this disease.

Pamela reports a positive relationship with the doctors and noted that their caring approach extended into their personal life when the doctors attended Ryan's high school graduation party.

FAMILY ROUTINE AND ROLES

Pamela said that Ryan's uncommon method of diagnosis did not create a family reaction and did not change family roles. Their large extended family has been supportive and helps taking care of the other children during medical admissions. Now that their children are older it has become easier for caretaking during medical crisis. Pamela and her husband split time between work and being at the hospital to minimize the effect on their jobs and family. She stated:

Hmm, because of how it presented itself, it wasn't a big deal. He's had it for all these years and there have been no consequences so it's always been business as usual. Nope. That's one of the things about it and I think that's why Ryan's been so successful with this disease is because we changed nothing.

Pamela and her husband have consistent rules and high expectations for all their children. Pamela felt a stable routine allowed her family to react to the disease without changing their way of life. She reported:

You know the biggest thing that I can tell any parent with the sickle cell disease is don't treat him any differently. You have to react to the disease and the situations that present itself but never treat...he got in trouble like everybody else. He got on punishment like everybody else. He had chores like everybody else. And, I think that's helped him...that his expectations were the same as the other two children.

She felt that consistent expectations reminded Ryan that he is normal and increased his ability to exceed in life and in school. She encouraged independence for all her children and felt that school was part of their job during childhood. Pamela said:

You know what? The only time he got any slack was when he was in crisis and wasn't feeling good. And his sisters picked up the slack for him. So it was business as usual...it's always been business as usual for Ryan. I have never coddled Ryan. He went to high school. I never told him he could be homeschooled...he didn't have to go to

school...he always had to go to school...he always had to do well in school...his job was to do well in school and that he needed to be able to take care of himself once he gets to be an adult. That he couldn't rely on anybody but himself.

Pamela's positive messages of independence have helped Ryan developmentally transition into adulthood and attend a state university. She reflected that Ryan would need to independently take care of his body and the disease in the future.

SUMMARY

According to Pamela's reflection, Ryan's diagnosis was unusual and uneventful. His medical complications are minimal, and he is thriving in college. Pamela shared little remorse or worry about how sickle cell disease has affected their family over the years. She has high expectations for all her children so that they can be independent adults. Pamela approached this interview with hesitation but noted at the conclusion that it was not as difficult to reflect as she thought. Her interview style was no nonsense, which appears to coincide with her parenting style of a child with a chronic illness. She spent less than twenty minutes with the interview and then promptly returned to her child's bedside.

CAREGIVER TWO: KATHY

Kathy is a forty four year old mother of three children ages twenty four, thirteen, and ten years old. Her oldest child lives independently outside the home and her youngest child Ben has sickle cell disease. Kathy is single and is unemployed though actively seeks employment. She attended college but was unable to finish. She considers her educational attainment to be high school level. Kathy states her income level is below \$15,000 a year. She relies on government food stamps and the Supplemental Security Income payments she receives for Ben's disease. The interview with Kathy was conducted at Hurley Medical Center Sickle Cell Clinic during Ben's regularly scheduled exchange transfusion appointment.

THEMES

KNOWLEDGE OF DISEASE

Kathy recalls learning about sickle cell disease while visiting in Michigan when she was pregnant with Ben. During a regularly scheduled doctor's appointment, Kathy learned about sickle cell anemia testing for newborns in the State of Michigan. Kathy was aware she carried the sickle cell trait but was unaware of what the diagnosis of sickle cell disease would mean for her child. She stated:

Ben was born December 6. His first 2 weeks at home was pretty bad. He was always sick every day, we took him to the hospital, no one knew what was wrong with him or that he was even tested positive for sickle cell. By December the 24th, the day before Christmas, someone from the Sickle Cell Association of America called me, the Director explained to that she was going on vacation and that my son had been tested positive for sickle cell disease. That was left on my answering machine. So I was kind of like lost.

Kathy tried to gather information about sickle cell disease while caring for her sick newborn. She remembers increasing frustration with the medical providers. She felt lost and unable to care for her child.

Ben also had complications from his sickle cell disease which has affected his physical and cognitive abilities. She noted:

Before he was 3 years old he had some transducers and they said that they were abnormal. So before age 6, by the time he turned 4, prior to that before the strokes, he just stayed in the hospital constantly. But back to when he was 4 years old – before he had his first mild stroke – I didn't know anything about reactions to strokes or anything like that – the symptoms. He had a stroke at home and I let him go back to bed because I thought he wasn't feeling good. And, they always say check him at home first before you call the doctor. The next morning he didn't get up so that's how I knew he was really, really sick cause he didn't get up out of the bed. So I took him to the hospital and they said he had possibly had a stroke. He was kind of impaired. He had a stroke on his right side – so he's impaired on the left. He went through some therapy and by age 5 he had a second one. That legally blinded him in his right eye, he's still impaired in the left side because it was on his right side.

Kathy states she has educated herself on sickle cell disease but feels that others still do not understand. She reflected:

When you try to explain it to them they don't take it as seriously as you can take it because they don't know the symptoms like you know them. Being overworked – that's a problem. Being a heavy heat – that's a problem. Not having the proper nourishment – that's a problem. Not having the proper medications – that's a problem. So that's a lot of things that you have to follow up with to stay healthy when you have a sickle cell. The actual disease. Ben is SS. There are 4 different kinds: SC, Thalassemia and infant sickle cell disease. Ben has the worst in the world – which is SS. That's why he's on the transitional program which he got on in 2005 after he had his second stroke until now which is 2011. He started out every 2 weeks on the transitional program getting a transfusion. Now he comes every 4 weeks.

Ben receives medical care at multiple hospitals dependent upon his medical needs. Kathy has taken him to C.S. Mott Children's Hospital, Hurley Medical Center, and Covenant Hospital. When asked about her feelings towards the medical team, Kathy simply replied:

They keep him alive. He ain't gone yet. They keep him alive.

RESOURCES AND SUPPORT SYSTEMS

Kathy reports a lack of resources and support for her family. She struggles financially and relies upon government assistance. Her ability to work is affected by the need to constantly care for her sick child. Kathy's mother was her main support person but recently died from cancer. She noted:

Yes, ma'am. We have lack of transportation. So many means you have a serious problem. The last 2 years I haven't worked because Ben was pretty sick and also my mom had cancer. She's deceased now and it has been a very serious struggle. 'Cause there's no means for help for you out there. I can't apply for FIA and sit at the FIA building because if you miss a class you don't get your grant, so all I can really apply for is food stamps. I do get SSI for him which is \$600....

Kathy reflected back on the many factors of having a child with sickle cell disease and the resources needed. She recalled:

Early education on, paying attention to different little things that you probably wouldn't think you were paying attention to that you should pay attention to with someone who has sickle cell. Their environment status, your living. I've moved so many times, being involved with protective service cause of unstable environment of living. It's just so much you have to have straightened out and when you don't have the means for it all at once it's a bother. But you just keep trying to push on to do better. It seems like it gets worse and worse. But I'm at the standpoint now where things are better for me. I just need a job. I can handle the sickle cell disease much better than what I could in the beginning.

Ben's medical complications have also affected his ability to participate in school. Kathy is making Ben's education her current top priority as he is lagging behind his classmates. She stated:

He had hyper attention problems – he always had ADHD – he always had crises. He never had a normal childhood to grow and go to elementary school. His learning ability is very low. He needs the medicine seriously for ADHD but they say they can't give him any right now due to they counter-react to the medications that he on for iron deficiency.

Kathy has had to advocate for Ben to be meet the requirements for special education.

This constant need for advocating has also affected her job outcomes. Kathy noted:

His education is my main concern right now because he can't read, his adding is off, his penmanship, he still writes like a kindergartener and he's in the 5th grade. He has been ... 2 of the things ... the board of education say different things to you about how they can help you but it's not true. I had to fight for him to get him to get him in special ed and that took 3 years ... this is the second year of special education. So, I don't know how they can just set a child in a classroom and say that he can keep up with the rest of the kids when they don't even know the first word on the page. And, that's how I lost a lot of my jobs. His ability to sit down and maintain his behavior in the classroom so that was a problem. So, I'm at the school building every day, getting called from work...you can't get called from work like that.

Kathy finds help in the Sickle Cell Association of America. She tries to attend local support group meeting but finds attendance is low and families unwilling to connect. She noted that C.S. Mott Children's Hospital has a highly successful support group with built in supports during crisis. She commented:

I think that Ann Arbor has the best support group, though. They have a lot of families down there and they do different things with the kids. You have a buddy if your child goes in the hospital to support you while your child is there so it won't be so stressful. That buddy can come up to the hospital and sit with you or if you just need a break to get away, that buddy can sit there for you. In my situation, I never get a chance to leave the hospital from the day he get admitted to the day he leaves. It's like I always have to have a bag ready – like I'm still pregnant. Like I'm still pregnant.

Kathy also finds support in her faith in God. She feels that not all church parishioners are supportive of her decisions for Ben, but finds solace in talking with God. She stated:

Surprise, really, I'm not on drugs I really am. I look at a lot of the other people that come to the support group they talk about that. They get on drugs, turn into alcoholics. It's just a crazy mind sometimes. But, I read the Bible a lot. I just test it to myself a lot. I really do. Yes. I talk to God a lot. I talk to my daughter. 'Cause nobody else really appreciates your problem.

FAMILY ROUTINE AND ROLES

The family routine is day to day dependent upon Ben's needs. When asked about good days and bad days at their house, Kathy replied:

No we just going on a wishing well. Whoever gets up first checks Ben. He's usually up before everybody. But I know when he's sick he just sit down, he isn't gonna move. He's mostly energetic so when he ain't moving something's wrong. Something is really wrong.

Kathy knows sickle cell disease has been hard on her other children as her attention is always focused on Ben. Kathy reflects that her children have never spent a night without her. Her loss of independence after becoming a parent is overwhelming because of lack of family support and outside resources. She recalled:

You never get a free day after you have kids. My kids have never been away from me since they been born in the world. Ben don't stay all night nowhere, so Molly choose not to stay 'cause Ben can't.

Sarah, Kathy's eldest daughter, lives nearby with her own children. Kathy tries not to burden Sarah but relies upon her to care for Molly during long hospital admissions. Kathy stated:

But my daughter, she do pretty good. She have 2 children of her own and I just hate to put the burden on her too because she need to live her life too. And it do stop your whole world. It really does.

Ben's sister Molly comes to clinic appointments because she cannot be left home alone during all day appointments. Molly also does yearly book reports about sickle cell disease so she can educate her fellow classmates. Kathy says Molly and Ben are inseparable unless Ben is hospitalized. Kathy noted:

So Molly is like Ben's best friend. He don't get out of eyesight from her. She's in 8th grade and he's in the 5th. She goes to a Catholic school but she would like her brother to come to school with her but they don't have special education there. So they could be closer to each other. So that is a concern for her. She don't do too many activities outside, either because she know he can't play outside and do much running due to his port.

Kathy also struggles to discipline Ben because of his sickle cell disease and because of her past involvement with Child Protective Services. She feels Ben already carries the burden of sickle cell disease and could not tolerate punishment. She commented:

They say you can discipline someone with sickle cell but I don't see how you are going to chastise a person who has to suffer like that. I know you want them to learn to be good, to be obedient, to be truthful, trustworthy, respectful ... but when he do things wrong I don't spank him. I don't spank Ben. I talk to him ... I try to talk to him to the best of my ability. Because if he get hit, pushed, shoved – he could swell up. That's a problem with Child Protective Service even though you did spank him and you discipline him, they not understanding that. They see that as neglect. You do what really is the best you can. You going to protect your own child. I know I would never hurt mine. I love mine too much. I really do. I'd give up my own life for my children. And they know that. I would not eat for them to eat. They know that.

When asked about hopes and dreams for the future, Kathy's response was honest. She just tries to get by one day at a time because her experience with sickle cell disease has made life seem abnormal. She noted:

I just worry about day-to-day to look for hoping and dreaming is gone cause they just don't care for sickle cell and I don't see it as a getting better thing I just hope I live to see Ben grow up. I really do. I just want him to be able to grow up and take care of himself. Not depend on nobody.

Kathy spoke of being emotionally overwhelmed and finding support in her sick child.

Kathy recalls Ben telling her not to cry which led to greater feelings of personal loss and sadness.

She recalled:

But, one thing I can say about him is he ain't never, never cried about his problem. I don't know if he don't fully understand it yet, which I've explained to him on several occasions, but he never cry. He never cry when he go to the hospital. He never cry. He always try to get up to be strong. I cry more for him than he cry for himself. I can say that. And I don't know that if it's my tears that keep him from crying. He can't have surgeries or go to the doctor appointments and be at home sick. He just try to go on. He really do. When I was there, he tried to move or do something just to make himself happy. So he is very strong through it. He better than me. I can say that – he better than me. Sometimes you feel like you lose yourself.

Kathy's talked about her feelings of being lost because of her sick child. Kathy wondered how other parents get through life with a child with cognitive delays. She noted:

So you're really a nowhere case when you have a sick child. I really have always wondered how parents really function with a child that was retarded, though. Even though my son doesn't have a full disability, just for him to lack has been a problem for me. So I really, really, really don't understand how anybody who has a child that's retarded is making it. And that's for real. It's always been on my mind, it really has.

SUMMARY

Kathy began to gain knowledge of sickle cell after a traumatic first few months of Ben's life. Her child has a complicated medical history and requires a lot of daily care and management. Her relationship with the medical team is complex and spans three different hospitals and medical teams. She openly describes the emotional and physical burden having a sick child has had on herself and her family members. She has had to adapt her parenting style because of the burden sickle cell disease already places on Ben. Her family and community supports are limited, and resources are extremely scarce. Kathy's current focus is on improving Ben's education and finding employment so she can support her family.

CHAPTER FIVE

DISCUSSION

INTRODUCTION

The purpose of this study was to investigate the affects of chronic sorrow and illness ambiguity in caregivers of children with sickle cell disease. Two caregivers were interviewed at Hurley Medical Center, and their stories present a strong contrast of how illness can affect the child, the family, and the family's adaptation to illness.

KEY THEMES

KNOWLEDGE OF DISEASE

Pamela's child was not diagnosed until age eight and was hospitalized approximately one time per year. This atypical diagnosis helped maintain Pamela's belief that her child was normal and that sickle cell disease would only be a small part of his life. Pamela immediately began buffering new stressors at the time of diagnosis with existing resources. She developed a trusting relationship with Ryan's doctors early on and sought out additional information about the disease as she needed to process new symptoms or hospitalizations. She acknowledged unique features about the disease and her child but relied on the medical team for explanation and to develop a new care plan as needed. From age eight to currently at age nineteen, Ryan has had a consistent set of doctors. This consistency encouraged Pamela to develop a relationship and trust the medical team during times of crisis.

The FAAR Model shows that developing balance between stressors and resources enables restructuring and eventual family adaptation. The temporary need for adjustment allows the family to maintain current roles while planning for future restructuring. Meaning of the

hardship must be developed by all members of the family, and it may change over time (McCubbin & Patterson, 1983).

In contrast, Kathy's child was diagnosed through newborn blood screening required in the state of Michigan. Kathy knew she was a sickle cell trait carrier but did not understand what sickle cell disease was. Her previous two children did not have the disease, so she did not understand why Ben was sick in the first few weeks of life. Kathy received the diagnosis around Christmas after receiving a message on her answering machine. Kathy sought out medical care immediately but was provided with few answers in the first few weeks of Ben's life. This traumatic diagnosis likely created feelings of distrust between Kathy and the medical team. She acknowledged that her child receives medical care at three different hospitals under three different medical teams. Conflicting information between the hospitals likely added to Kathy's confusion about the medical care needed for her child. Ben has had many complications from sickle cell disease, including multiple strokes, vision impairment, and cognitive delays affecting his school ability. His medical care is complicated and often requires weekly doctor's appointments as well as frequent hospitalizations at any of the three hospitals where Ben receives care. Kathy's experience echoes research by Tomlinson and Harbaugh (2004). Her inability to get accurate information about her child's illness and prognosis increases uncertainty and undermines her confidence as a mother and caretaker. Family adaptation is affected by this uncertainty (Tomlinson & Harbaugh, 2004).

Kathy immediately developed a negative meaning towards sickle cell, and this confusion surrounding the disease has continued with three hospitals and three medical teams. Her feelings towards the medical team are basically that they keep her child alive. Ben has many complications from sickle cell disease which also make the disease seem more of a burden. Her

adjustment periods with sickle cell disease are continual as he faces new medical complications. She has little ability to complete the FAAR cycle because of failings at the initial stages of family adjustment to sickle cell disease. Kathy's continual struggle to restructure has likely led to the stage of exhaustion in the FAAR Model. Due to limited resources and unsuccessful attempts at restructuring, she has exited the cycle and stopped at exhaustion (McCubbin & Patterson, 1983).

FAMILY ROUTINE AND ROLES

Pamela did not allow sickle cell disease to become a dominant feature in their family life. She attributes much of this to Ryan's delayed diagnosis. Family expectations were the same for all three children, and allowances were made only temporarily if Ryan was sick. Pamela felt long term goals for her children were important, and Ryan was expected to learn to take care of himself. Ryan also was expected to succeed in school, which encouraged feelings of normalcy with his peers. Ryan was encouraged to take over his medical care during the teenage years, which increased his ownership over the disease and feelings of normalcy. Ryan also was adept at caring for his medical needs by the time he left for college.

Pamela's feelings of independence for each child may have minimized the stress of illness on the greater family. Pamela did not reference the unknown of Ryan's future with sickle cell disease during the interview. In fact, she was confident of his future because of their positive family values. Pamela's family values encouraged an internal locus of control. Research by Barakat, Lutz, Nicolaou, and Lash (2005) found that parents with an internal locus of control exhibited more self competence and self esteem while caring for their child (Barakat, Lutz, Nicolaou, & Lash, 2005).

Medical teams often encourage patient independence during the teenage years to teach caretaking routines previously performed by parents. Medical adherence can drastically change during late teenage years if parents do not teach independence. Pamela taught Ryan that sickle cell disease would not dominate his life and that he already had the life skills to be independent. Life Course Theory and the FAAR Model both address meaning making. Pamela believed sickle cell disease would not overwhelm family life and early on began the steps for family adaptation. Life Course Theory notes that the family and its needs change over time. Before and after the diagnosis of sickle cell disease, Pamela and her husband maintained a stable lifestyle for their children. The family was financially stable with two working parents. Stable finances were a major resource that helped buffer additional stressors in their family life. Ryan's medical needs changed as he grew older, but the family's values remained consistent. Pamela's parenting style of independence became even more advantageous as Ryan grew older. Pamela did not reference any feelings of loss for herself or her child. Her early restructuring and adaptation of the family routine and roles may have buffered any feelings of loss. Ryan's sickle cell disease was well managed through childhood and into adulthood, which likely minimized feelings of loss.

Kathy's beliefs about sickle cell disease are in strong contrast to Pamela. Kathy believes that sickle cell disease plays a dominant role in their daily life, and their household routine depends upon Ben's needs. They have no long term family goals, but instead struggle with meeting day to day needs. Kathy often spends part of each day at Ben's school advocating for his educational needs, as he is significantly behind his peers. Multiple strokes have left Ben cognitively impaired. Kathy noted that no one at school advocates for her child, so she must sacrifice her day to keep Ben in school. She is also often called to school due to Ben's behavior issues in the classroom. Research by Canam (1993) suggested that discipline reminds the ill child

of limits and helps to normalize family life. The ill child is treated with the same expectations as the well siblings (Canam, 1993). The interview context suggests Kathy does not believe in use of discipline for Ben. She feels his burden with sickle cell disease is punishment enough.

Kathy's perception of sickle cell disease was negative since diagnosis. The meaning she has attached to this stressor is that the disease is a burden. The FAAR Model suggests that the meaning attached has affected her ability to restructure and adapt her family's lifestyle. Kathy is continually trying to adjust to sickle cell instead of moving forward towards restructuring.

Kathy acknowledged that she feels lost and burdened by sickle cell disease but feels that is her burden to carry. She has no personal time and has never left her children alone with another caregiver. Kathy's children are all still dependent upon her for daily care. If Ben is hospitalized, Kathy remains at the bedside for the entire hospital admission. Kathy's oldest daughter lives independently with her own children, but is often asked to take on the responsibilities of her younger sister Molly. Ben's sisters help provide daily care and adapt their work and school routine based on whether Ben is well or ill. Molly often misses school because she accompanies Kathy and Ben for outpatient clinic appointments. Kathy is unwilling to send Molly to school and have her get home independently due to the poor neighborhood in which they live. The drive to the hospital is lengthy, and appointments last several hours.

Ben's needs set precedence over the needs of the entire family. Their lack of daily and long term goals has created a fatalistic attitude towards the disease. Kathy feels she is independently carrying the burden of disease for the whole family, but in reality they are all suffering the effects of Ben's sickle cell disease. Mu and Tomlinson (1997) indicated that parents who remain uncertain with their child's illness and prognosis will face increased ambiguity and

parental distress. Research suggested caregivers learn to self regulate their family system quickly during crisis to minimize the negative stress (Mu & Tomlinson, 1997).

Kathy's acknowledgement of feelings of loss and burden allude to feelings of ambiguous loss and likely chronic sorrow. The family roles are dependent upon the needs of Ben's sickle cell, and boundary ambiguity fluctuates when Ben is hospitalized. The unknown of Ben's future with sickle cell disease weighs heavily on the family.

RESOURCES AND SUPPORT SYSTEMS

Pamela's family had a strong set of resources and family supports which likely minimized the effects of sickle cell disease. Pamela is married, and she and her husband are both college graduates. Their understanding of sickle cell disease was supported by their level of educational attainment. She acknowledged seeking out the medical team when she had questions. Her spouse is her main support person, and they are able to divide parenting and individual responsibilities during times of good health or crisis. Their employers were flexible with time off likely because Ryan's hospitalizations were infrequent. Their employment was not at risk of termination, which eased the stress during Ryan's illness. Their annual income exceeds \$65,000, which allowed for financial stability and would allow for time off even if it was without pay. The expense of Ryan's medications, appointments, and hospitalizations would be buffered by the private insurance offered through an employer as well as Children's Special Health Care Services which provides additional coverage for children with sickle cell disease.

Pamela and her husband also have a large extended family that lived locally and was available if the other children needed support during Ryan's hospitalizations. Pamela only utilized extended family supports temporarily during a crisis and then reverted back to the roles in the immediate family. Her extended family likely did not feel burdened by the disease because

their stress was only temporary. It also would make them more likely to help out again during crisis. Pamela was using temporary restructuring during crisis as a step towards family adaptation. As her children grew older and began new life phases, their burden on extended family during crisis decreased over time.

Kathy's lack of supports and resources has amplified the burden of her child's sickle cell disease. Kathy is unemployed and relies on government assistance. Her struggle to get off government assistance is devastated by Ben's constant medical complications or behavior issues at school. Since Kathy's mother passed, her oldest daughter has become her primary source of support. This support is limited as the daughter has her own children. Kathy acknowledges her strong faith in God has helped her maintain her sanity during the years of medical complications. However her faith does not help fix her issues with unemployment, Ben's medical complications, or issues at school.

The FAAR Model highlights that a lack of resources makes buffering new stressors difficult. Kathy's stressors often outweigh her resources which does not allow for development of coping strategies. Kathy copes by just getting by day after day. Ben's health does not remain stable long enough for the family to complete the adjustment, restructuring, and adaptation cycle.

Ben's medical needs also have remained high over several life stages. This has not allowed Kathy and the family to gain control over sickle cell disease. As Ben has grown older, he has not gained independence like his peers due to his environment and disease. Ben's siblings also have not gained as much independence as their peers because of the need to care for Ben. Kathy spoke of never having spent a night away from her children and being their sole caretaker. The micro environment in which Kathy lives also has hindered her abilities to adapt to the disease. Her low socioeconomic status makes existing and new stressors more taxing. She admits

to living in a poor neighborhood, child protective services involvement, and chronic unemployment.

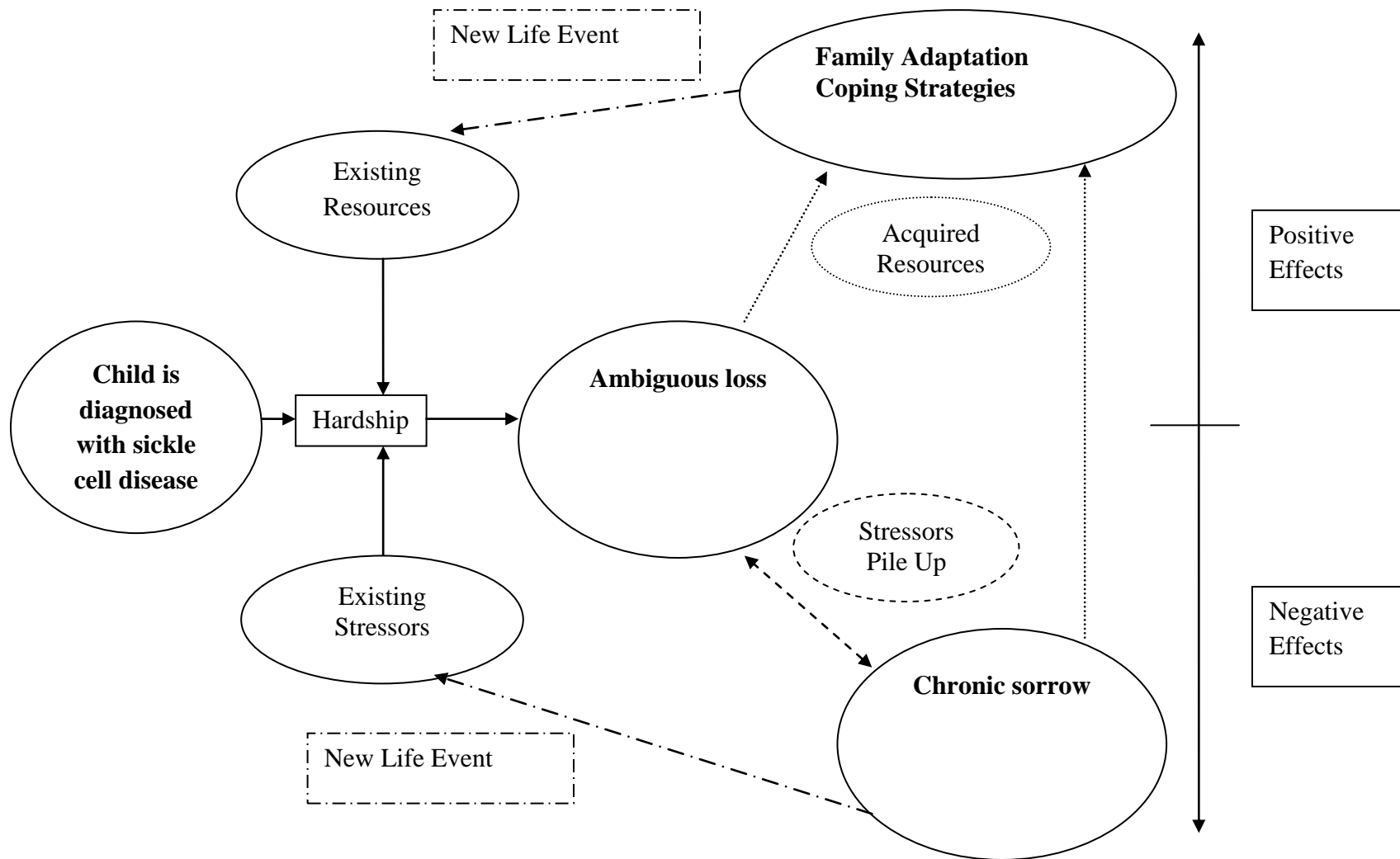
SUMMARY

Pamela and Kathy represented two different approaches to caring for a child with sickle cell disease. While they share the same medical team and live in a similar geographic location, their pathway through the experience is dramatically different. Pamela appraised her family resources and stressors and attached meaning to her child's diagnosis early in the process. She developed a relationship with the medical team which buffered new stressors when additional hardships occurred. Her feelings of ambiguous loss were minimized as her family restructured, developed coping strategies, and adapted to sickle cell disease. Pamela recognized the need to flex family routines and roles temporarily when Ryan was sick, but quickly moved her family back towards coping and adaptation.

Kathy's pathway shows the negative repercussions when family adaptation does not occur. Her lack of resources is not only an initial problem but amplifies as additional stressors occur throughout her child's lifetime. The meaning attached to her child having sickle cell is negative and burdensome. Kathy noted feelings of loss due to confusion about sickle cell disease and its outcome for her child. These continual feelings of ambiguous loss suggest evidence of chronic sorrow. Kathy's day to day coping strategy makes future family restructuring and adaptation more difficult.

FIGURE 2

REVISED MODEL OF SICKLE CELL DISEASE AND CHRONIC SORROW



REFLECTION ON THE CHOSEN METHODOLOGY

The qualitative case study approach to this study was beneficial to discover resource and stressor themes in each family. The two caregivers interviewed created a contrast between case studies. The *Burke/NCRCs Chronic Sorrow Questionnaire* was used as a reference point to develop interview questions. This validity and reliability of this questionnaire had already been tested in a Mary Burke study in 1989 (Burke, Hainsworth, Eakes, & Lindgren, 1992). The addition of open ended interview questions allowed the caregivers to reflect on details of their experience with sickle cell disease that may not have arisen with a formal questionnaire. One caregiver noted that this interview was easier than she expected. The other caregiver had a very emotional interview that involved several breaks to check on her child. This caregiver also asked for the tape to be stopped because she was too overwhelmed to reflect anymore. For one caregiver, interviewing in proximity to her child was important.

Had I completed more interviews, I may have been able to ascertain whether interviewing during inpatient hospitalization versus outpatient appointment had an effect on caregiver response. I think qualitative research similar to this helps to identify family needs and reactions to a chronic disease. As the caregiver's interviews showed, it appears some families succeed in the face of chronic illness and others struggle year after year.

There was a benefit of being a member of the Hurley Medical Center team. This gave me credibility as a researcher and it gave confidence to the caregivers that someone was listening to their story. While I was not a direct member of their medical team, I was not an outsider prying into their lives. There is a strong sense of community within Flint. Parents often note that people outside of Flint do not understand the additional stressors they face. I think it also gave hope that future support was possible if their story was heard. Parents consistently note the lack of support

and lack of communication with the medical team surrounding the needs of their child and family. One parent's concerns are often similar to that of another parent.

LIMITATIONS

There are several limitations of this case study that are important to note. The study involved only two caregivers. This gives only a brief glimpse into the lives of caregivers of children with sickle cell disease. The interview questions were open ended but did not allow for follow up questions. Certain caregiver responses hinted at the need for additional follow up questions, but the interview protocol did not allow for that. The two caregivers presented a contrast of views towards their child and sickle cell disease. These caregivers also represented very different socioeconomic status, which likely affected their views of sickle cell disease and ability to utilize resources. The caregivers also were interviewed while their child was experiencing differing situations. One child was having an outpatient clinic appointment and the other child was sick and hospitalized inpatient during crisis. A different methodology might identify more similarities or differences between caregivers.

Hurley Medical Center has a small population of children with sickle cell disease that met study criteria. Other hospitals with larger populations would likely find more willing participants. There also was an interruption during recruitment due to the principal investigator taking an extended leave due to family emergency. This likely decreased interest in the study and possible caregivers also may have been missed during this period of time. The principal investigator also was not the direct source for caregiver recruitment. Members of the Hurley Pediatric Sickle Cell clinic handed families a letter detailing the study. The delivery of the study information may have affected whether caregivers were willing to participate.

FUTURE RECOMMENDATIONS

A larger study is needed to identify specific areas that the medical team can improve upon support for caregivers. Studying caregivers longitudinally would help identify family stressors along the child's life course. Support also could be developed if anticipatory stressors could be identified. Hurley Medical Center has no formal support groups or services offered to families of children with sickle cell. One caregiver that was interviewed suggested a buddy system modeled after a program in another hospital. Caregiver feedback before and after new support services are offered could prove beneficial. Feedback from the medical team about their view on family resources, stressors and adaptation might highlight a dissonance between the family and the medical team.

Future studies also should investigate themes of resilience. The caregiver interviews hint that resilience could be a factor in whether families succeed or fail when faced with chronic illness. A study analyzing factors that buffer family stress and resistance may highlight areas for medical team improvement.

IMPLICATIONS

Children with chronic illness are now living into adulthood because of advances in medical technology. Due to a change in care perspective, the medical team is now concerned with how illness affects the family as individuals and as a whole. Family centered care initiatives encourage support for the whole family, which means better understanding of factors in family success as well as challenges with chronic illness.

Sickle cell disease requires extensive daily care, which changes roles and routines within the family. These contrasting case studies showed the differences in family roles and routines when managing the disease. Caregiver, Pamela, remained consistent with family expectations

and did not allow sickle cell disease to overwhelm their lives. Research by Canam (1993) highlights the importance of normalization of family routine within the family (Canam, 1993). Pamela also used problem solving coping strategies during times of medical crisis. Families must utilize a variety of resources to buffer the stressors of chronic illness. These resources may include finances, extended family, school counselors, and church family.

The caregiver contrast also highlighted a difference in coping strategies. Additional stressors such as lack of resources no doubt affect the family's ability to adapt. Financial instability is a major stressor that affects the ability to gain other resources. This finding aligns with research by Burnes, Antle, William, and Cook (2008). Mothers reported overwhelming feelings of loss when their social supports and resources were low. Caregivers of children with sickle cell disease noted increased feelings of social isolation and separation anxiety from other family members (Burnes, Antle, Williams, & Cook, 2008). However, teaching coping strategies can help buffer these stressors over which the medical team has little control. Positive appraisal of the disease and problem solving focused strategies has shown to assist with adaptation. Research by Ahmann and Rollins (2005) affirmed the need for caregiver coping strategies. Anticipatory guidance was found to decrease the pile up of stressors during crisis (Ahmann & Rollins, 2005).

It is advantageous for medical teams to encourage positive coping strategies and family adaptation. As these children with sickle cell grow into adults, they will be more likely to be independent and better manage their healthcare needs. This could decrease their dependence on the healthcare system and increase their life expectancy. Independent and healthier adults with sickle cell disease are more likely to go to college, find employment, and be successful in adulthood.

RESEARCHER REFLECTIONS

This thesis has been a lengthy process spanning several years of my career at Hurley Medical Center. I had the privilege of providing support to many parents of children with chronic illness and continually saw parents who struggled and those who succeeded. Limited resources drastically affect the caregiver's ability to cope and adjust to chronic illness. Some chronic illnesses have far more resources available to families than others. Caregivers note this imbalance in resources and have remarked whether it is better to deal with one disease or their other. I will soon be leaving Hurley Medical Center but still feel that caregivers of children with sickle cell disease require more support than currently provided. Local and national support and resources are limited for families with sickle cell disease. It is a heavy burden to carry as an individual let alone the effects on the whole family.

My experience with chronically ill infants in the neonatal intensive care has shown me the strength of a consistent medical team. Parents who developed confidence to ask questions in turn became more confident carrying for their baby. Periods of uncertainty and high stress were buffered by the strength of the relationship with the medical team.

Finally I reflect on my experience caring for my dad while he was fighting cancer. While his illness was not chronic, it changed our family and forced us to go through temporary restructuring and adaptation. Each member of my family had to individually assess his/her own resources and personal stressors. We each viewed this hardship differently and made temporary changes to accommodate the new demands illness placed on our lives. Boundary ambiguity influenced the family as we were used to living independently outside the home, and now were brought back together. We were often unsure of our roles as adult children and sometime medical caretakers. We could not conceive of being a family without my dad being there, but we faced the ambiguity. Family restructuring and adaptation has now cycled through several times

in the two years since my dad has passed away. We are a different family because of my dad's illness, but we are working towards becoming a cohesive family unit again.

APPENDICES

APPENDIX A: EMAIL TO SICKLE CELL CLINIC STAFF

April 16, 2011

Dear Hurley Sickle Cell Clinic Staff,

I am conducting a research study to investigate the lived experiences of primary caregivers of children with sickle cell disease. This study will explore the burden of chronic illness on the family as well as ways the medical team can provide support. After obtaining IRB approval, I will be recruiting three primary caregivers of children who participate in exchange transfusion clinic. Caregivers will be given an informational letter prior to interviews being conducted and they will contact me if they wish to participate. The interviews will be conducted in the clinic conference room during an exchange transfusion appointment. If I am unable to recruit three caregivers from clinic, I will recruit a caregiver from a general hospital admission. Caregivers will be required to sign a consent form to participate.

I look forward to working with you. If you have any questions, please contact me.

Sincerely,

Christine Bennett, CCLS
Hurley Medical Center
Child Life Specialist
810-262-9577

APPENDIX B: CAREGIVER INFORMATIONAL LETTER

April 16, 2011

Dear Primary Caregiver of a Child with Sickle Cell Disease,

My name is Christine Bennett. I am a Certified Child Life Specialist at Hurley Medical Center. In addition to working at Hurley, I am also a master's student at Michigan State University. I am interested in learning more about how pediatric sickle cell disease affects the family. This research is important so that the medical team can learn more about how to best provide support to your family and other families who receive care at Hurley Medical Center.

I would like to invite you to tell your story as the parent of a child with sickle cell disease. This is a research study. The purpose of this letter is to give you the information you will need to help you decide whether you want to participate or not. Participation in the study is completely voluntary. If you choose to participate, I would like to talk with you in person during your child's regularly scheduled exchange transfusion appointment. If you wish to participate, but are unable to come to clinic, please contact me so we can make other arrangements. Interviews will last no longer than ninety minutes.

If you choose to participate, I will provide you with a consent form to sign that will give further details of the study. Your name, family members' names, and any other identifying information will be kept confidential.

If you would like to participate, please contact me by phone or email. My phone number is 810-262-9577 and email address is cbenne1@hurleymc.com.

Thank you in advance for your help.

Christine Bennett
Child Life Specialist
Hurley Medical Center

APPENDIX C: HURLEY INFORMED CONSENT FORM

HURLEY MEDICAL CENTER

PARTICIPANT CONSENT FORM

Chronic Sorrow and Illness Ambiguity in Caregivers of Children with Sickle Cell Disease

Investigators:

Primary Investigator: Christine Bennett Certified Child Life Specialist Hurley Medical Center
Child Life Services One Hurley Plaza Flint, MI 48503 810 262-9577

Secondary Investigator: Dr. Barbara Ames, Graduate Program Director of Human Development
and Family Studies at Michigan State University 517-432-3324

Sponsor: No Funding Sources

I am asking you to be in a research study. This study is being conducted jointly by Michigan State University and Hurley Medical Center. The goal of this consent form is to give you the information you will need to help you decide whether or not to be in this study. Please read the form carefully. I can help you read and understand the form if necessary. You may ask questions about the purpose of the research, what I would ask you to do, the possible risks and benefits, your rights as a research participant, and anything else about the research or this form that is not clear. When all your questions have been answered to your satisfaction, you can decide if you want to be in the study or not. This process is called informed consent.

SITE OF THE RESEARCH STUDY

Where will the study be conducted?

Hurley Medical Center Pediatric Sickle Cell Clinic

PURPOSE OF STUDY

What is the purpose of the research?

This research study is being done to (1) learn about being a caregiver of a child with sickle cell disease (2) explore family resources and stressors (3) improve support services offered by the medical team.

Who is being asked to participate in this research study?

You are being asked to participate in this study because you are the primary caregiver of a child with sickle cell disease.

PROCEDURES

What procedures will be performed for research purposes?

Primary caregivers of children in exchange transfusion clinic will be given a letter with information about the research study. You will be interviewed during a Sickle Cell Clinic appointment. You may also delay the interview for one month if there is not enough time. Interviews will last about an hour and a half.

A caregiver from a general hospital admission will be invited to participate if there are not three caregivers from clinic. The caregiver from the general hospital admission will follow the same interview process as caregivers from Sickle Cell Clinic.

The interview style will be flexible and informal. I want to hear your story with sickle cell disease. Questions will explore family life, sources of stress and support, and experiences with the medical team. Questions will also explore your feelings and the feelings of other family

members. You may refuse to answer any question if you feel uncomfortable. Interviews will be audio recorded.

BENEFITS

What are the possible benefits to taking part in this research study?

There are no expected direct benefits to you for participating in this study. Information from this study may help other caregivers of children with sickle cell disease. This study may also help the medical team to understand how to help support caregivers.

RISKS

What are the possible risks, side effects, and discomforts of this research study?

There are no physical risks to participating in this study. There may be emotional risks because of the type of interview questions. You may feel uncomfortable as you talk about your family and sickle cell disease. You may feel concerned about your family's privacy as interview questions ask about other family member responses to sickle cell disease.

If you need to talk to someone following the interview, a referral can be made to the Michigan State University Couple and Family Therapy Clinic or to Genesee County Community Mental Health.

Will I be told of any new information or new risks that may be found during the course of this study?

Any new information gathered during this study that uncovers new risks will be given to you in writing. If new information is given, consent to participate in the study will be re-obtained.

ALTERNATIVE PROCEDURES

What treatments or procedures are available if I decide not to participate in this research study?

There are no other procedures or treatment associated with this research study.

CONFIDENTIALITY STATEMENT

All information connected with this study will be kept confidential. To maintain privacy, you will be interviewed in a private conference room. There will be no additional members of the research team or any staff members of Hurley Medical Center in the conference room during the interview. Information about you will be kept confidential to the full extent allowable by law unless there is a danger to yourself or others.

Information from the interviews will be used to complete the primary investigator's master's thesis requirements at Michigan State University. Information will be written out and audio tapes will be destroyed after completing the Michigan State University thesis requirements. To maintain the confidentiality of your records, the preliminary study information and study results will be stored in the primary investigator's locked file cabinet for three years after the closure of the thesis project. This file cabinet is located at the Michigan State University Department of Human Development and Family Studies 7 Human Ecology Building East Lansing, MI. Preliminary information and study results can only be accessed by the primary investigator Christine Bennett, the secondary investigator Dr. Barbara Ames, the Michigan State University Institutional Review Board and the Hurley Medical Center Institutional Review Board. The primary investigator's Michigan State University advisor will only review information after alias names have been applied.

Who will know about my participation in this research study?

A professional transcriptionist may be used to write out interview information. Audio tapes will be erased after data has been written out and reviewed. The primary investigator will be the only other person to listen to the audio tapes.

Staff members of the Hurley Medical Center Pediatric Sickle Cell Clinic will refer caregivers for study participation and help to schedule interviews. For that reason clinic staff members may be aware of your study participation.

The results of this study may be published or presented at professional meetings, but your identity will remain confidential through use of an alias.

COMPENSATION/INJURY CLAUSE

Will my insurance provider or I be charged for any costs of any procedures performed as part of this research study?

In case you need to talk to someone after the interview, a referral can be made to the Michigan State University Couple and Family Therapy Clinic or to Genesee County Community Mental Health. These therapy appointments are available at their usual cost. No money has been set aside for additional therapy needs. You or your insurance company will be charged for continuing therapy appointments. You will receive no payment for taking part in this study. You will be responsible for scheduling any therapy appointments.

Will I be paid for participating in this study?

There is no money provided for participation in this study.

VOLUNTARY PARTICIPATION AND WITHDRAWAL

Is my participation in this study voluntary?

Participation is voluntary, you may choose not to participate at all, or you may refuse to participate in certain procedures or answer certain questions or discontinue your participation at any time without consequence. If you refuse to participate, there will be no penalty or loss of benefits to which you are otherwise entitled. You have the right to leave the study at any time. If you leave the study, there will not be any penalty or loss of services to which you are otherwise entitled.

Can I be removed from the study without my consent?

The primary investigator may decide to remove caregivers participating in the study if the need arises. Removal of caregivers may occur to protect your health and safety.

CONTACT INFORMATION

Whom do I contact if I have any questions about the research, or if I'm injured while on the study?

If you have concerns or questions about this study, such as scientific issues, how to do any part of it, or to report an injury (i.e. physical, psychological, social, financial, or otherwise), please contact the researcher:

Christine Bennett

Hurley Medical Center

Child Life Services

One Hurley Plaza

Flint, MI 48503

cbennel@hurleymc.com

(810) 262-9577

Dr. Barbara Ames

Michigan State University

Human Development and Family Studies

7 Human Ecology Building

East Lansing, MI 48823

ames@msu.edu

517-432-3324

Whom do I contact if I have questions regarding my rights as a research participant?

If you have questions regarding your rights as a research subject, you can call the Hurley Institutional Review Board at (810) 262-9974 or by email at cadams2@hurleymc.com

Signature of Investigator

Date

SUBJECT'S STATEMENT

This study has been thoroughly explained to me. I volunteer to take part in this research. I have had the chance to ask questions and I feel my questions have been answered to my satisfaction. If I have follow up questions, or if I feel the need for additional support, I can call the primary investigator listed below. If I have questions about my rights as a research subject, I can call the Institutional Review Board at Hurley Medical Center at (810) 262-9974.

Video/Audio Tape Release

I give consent to be audio taped during this study:

Please initial: ____Yes ____No

Name of Subject (Print)

Signature of Subject/Parent/Legal Guardian

Date

Address of Subject/Parent/Legal Guardian

WITNESS SIGNATURE

My signature as a witness certifies that the subject signed this consent form in my presence as his/her voluntary act and deed.

Name of Witness (Print)

Signature of Witness

Date

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APPENDIX D: CAREGIVER INTERVIEW

This research study is being done to (1) learn about being a caregiver of a child with sickle cell disease (2) explore family resources and stressors (3) improve support services offered by the medical team.

All information connected with this study will be kept confidential. To maintain privacy, you will be interviewed in a private conference room. Information about you will be kept confidential to the full extent allowable by law unless there is a danger to yourself or others. Participation is voluntary, you may choose not to participate at all, or you may refuse to participate in certain procedures or answer certain questions or discontinue your participation at any time without consequence. If you refuse to participate, there will be no penalty or loss of benefits to which you are otherwise entitled. You have the right to leave the study at any time. If you leave the study, there will not be any penalty or loss of services to which you are otherwise entitled.

Thank you for telling your story as a caregiver of a child with sickle cell disease.

1. Tell me about yourself and your family.
2. Tell me about (blank), your child with sickle cell disease.
3. When did you first learn about your child's diagnosis?
 - a. How did you and your family react to the news about sickle cell disease?
 - b. How did you feel after receiving the new diagnosis?
 - c. What were your biggest worries when your child was first diagnosed?
4. In what ways did your family adapt?
5. What does a good day look like at your house?
6. What does a bad day look like at your house?
7. Do you have sources of support that have helped you?
 - a. Have these sources of support changed over the years?
8. What have been your challenges?
 - a. How did you respond to these challenges?
 - b. How did you respond immediately?
 - c. How did you respond over time?
9. Has your child been hospitalized before?
10. Did your feelings resurface or change after experiencing new challenges?
 - a. What helped you manage your emotions?
11. What have been your feelings about the medical team?
 - a. What was helpful?
 - b. What was not helpful?
12. What does sickle cell disease mean to you today?
13. What are your biggest worries today?
14. What do you hope for your child and family in the future?

There is also some general information about you and your family that I'd like to gather. Please fill out the enclosed form and place it in the envelope.

Age

Educational attainment

- Less than high school completion
- High school graduate
- Some college, no degree
- College graduate
- Vocational/Trade degree
- Master's degree/Doctoral degree

Marital Status

- Single
- Partnership
- Married
- Divorced
- Widowed

Occupation

Income level

- Under \$15,000
- \$15,000 to \$24,999
- \$25,000 to \$34,999
- \$35,000 to \$44,999
- \$45,000 to \$54,999
- \$55,000 to \$64,999
- Above \$65,000

Number of family members who live in the home

Number of family members with sickle cell disease who live in the home

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