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A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Family Caregivers

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A QUALITATIVE DESCRIPTIVE STUDY EXPLORING THE ADAPTATION OF FAMILIES OF CHILDREN WITH MULTIPLE SCLEROSIS FROM THE PERSPECTIVE OF CAREGIVERS

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ABSTRACT

Two to 5% of the US population of individuals afflicted with multiple sclerosis (N = 400,000) are diagnosed prior to the age of 18 with pediatric onset multiple sclerosis (POMS). Most children and teens with POMS have a relapsing remitting course of the disease with unpredictable relapse onset causing cognitive and physical disability. This can result in school absences, hospital and outpatient visits, poor academic performance and interruption in normal social activities. In turn, these individuals may develop an inability to provide self-care, which could lead to poor quality of life. POMS can affect the individual diagnosed but may also cause temporary and/or permanent disruption in the structure of the family. In the initial phases of the disease, families may adjust by making day-to-day changes to their roles and responsibilities. Over time, families may also adapt their level of functioning to meet the demands of the unexpected nature of the disease. No research exists on the factors that influence the adaptation of families of children diagnosed with POMS. The purpose of this qualitative descriptive study is to gather rich and in-depth accounts of the factor that influence how families adjust and adapt to diagnoses from the perspective of family caregivers.

Twenty female family caregivers who participated in a semi-structured, one-on-one interview with the researcher. The caregivers ranged in age from 28 to 55 years with a mean age of 44. The participants were caregivers of children that ranged from age 7 to

22 previously diagnosed with POMS for at least one year or more. In addition to the interview, each participant also provided socio-demographic data about themselves, the child living with POMS and other individuals living within the household. Verbatim transcripts were analyzed by thematic analysis using NVivo Pro software to organize the data into themes. Demographic data was analyzed using SPSS software. Seven themes emerged from the data: stress and strain; adjusting to the diagnosis; communication; coping with the diagnosis; sources of strength; achieving balance; and the overall experience of the family.

Overall findings provided insight into the family experiences of adapting to a diagnosis of POMS when there are unique challenges faced in this population. Findings also suggested implications for practice as well as implications for future research with regard to families of children with POMS.

Keywords: pediatric onset multiple sclerosis, qualitative descriptive, family adaptation, family adjustment

DEDICATION

First, I would like to dedicate this dissertation to all the families of children that have been diagnosed with demyelinating disorders that I have served at CPODD over the years. It has been an honor and a privilege to get to know you and to have an opportunity assist you and your families in the process of adapting to your new lives after being diagnosed with a rare chronic condition.

Secondly, I would like to dedicate this dissertation to my earthly father who is no longer with us in this realm but whose presence is always existent in my life. My father always instilled in me that I could do anything as long as I put my mind to it and stayed focused. I hope that I have made him proud.

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LIST OF ABBREVIATIONS

ADEM..... Acute Disseminating Encephalomyelitis

CNS..... Central Nervous System

CPODD..... Center for Pediatric Onset Demyelinating Disease

DMT.....Disease Modifying Therapy

FDA.....Food & Drug Administration

JIA..... Juvenile Idiopathic Arthritis

MRI.....Magnetic Resonance Imaging

MS..... Multiple Sclerosis

PML .....Progressive Multifocal Leukoencephalopathy

POMS.....Pediatric Onset Multiple Sclerosis

PPMS .....Primary Progressive Multiple Sclerosis

RAM .....Roy's Adaptation Model

RRMS ..... Relapsing Remitting Multiple Sclerosis

SPMS .....Secondary Progressive Multiple Sclerosis

CHAPTER 1

INTRODUCTION

Over the years, literature has accumulated with respect to chronic illnesses such as childhood cancer, diabetes, cystic fibrosis, sickle cell disease, asthma and the effects on the family of an affected child. Research has shown that the burden of having an chronic childhood illness within a family can produce strain in all domains of life: financial, physical, emotional, social and personal (Barlow & Ellard, 2005; Gold, Treadwell, Weissman & Vichinsky, 2011; Gustafsson, Olofsson, Anderson, Lindberg & Schollin, 2002; Williams et al., 2009). According to the United States Census Bureau, a family is defined for statistical purposes as "a group of two people or more related by birth, marriage, or adoption and residing together" (United States Census Bureau, 2011, B-4). Family members take on new and unexpected roles that may interrupt the usual daily routine and normal function of the household (Lawrence, 2012). Family activities such as keeping up with daily chores, schoolwork, jobs and social activities may pose an emotional strain on the family (Lawrence, 2012). Families may begin to feel socially isolated and alone from extended family and friends because others may not understand their experiences. Yet some may feel overwhelmed by extra attention received from extended family and friends because they have not had time to process their own personal feelings.

Financially, there may be unexpected medical bills not covered by insurance that may strain families. The lost time from work for the parents of the child with the chronic illness may cause a financial strain on a family because of the symptoms and the duration of time that these symptoms last. Payment for medications and co-payments for hospitalizations and outpatient treatments, such as physical and occupational therapy may prove to be costly to the family and add additional financial strain on the family.

Physical strain on parents may result from sleep deprivation due to direct care or emotional strain from worry and anxiety about the uncertainties of the disease. It may also result from the direct inability of parents to provide self-care due to the time demands required with the ill child and the demands of other responsibilities. In addition to the financial, physical and the emotional strain on the parents, there is an interruption in the social lives of all the other members of the family as well.

Younger siblings or children in the household may feel a sense of neglect, jealousy or abandonment when there is a child diagnosed with a chronic illness that requires a lot of parental attention and time. On the other hand, the older children in the household may have more responsibilities than usual when their younger sibling is chronically ill. Research shows that emotions and vulnerability are the most commonly cited effects documented on well siblings of a child with a chronic illness (Alderfer et al., 2010). At times of acute crisis, there may be a need for temporary living arrangements for the siblings or children living in the household if the child with illness requires hospitalization or extended treatment. During these times, spouses or other adults may feel neglected and children may feel jealous because the focus and attention is on the ill child.

Furthermore, the primary caregiver, the person who lives in the household and who assumes primary care and responsibility for the ill child, takes on the additional role of accompanying the child to the hospital or clinic for physical examination, consultation and treatment (Family Caregiver Alliance, 2017). In addition to the new duties assigned to the caregiver regarding the diagnosis, the primary caregiver must maintain his or her role as a parent and caregiver to the siblings of the ill child as well as a spouse and family member to others within and outside of the household. This may place demands on the caregiver not previously experienced. These demands may prove disruptive in the caregiver's daily professional work as well as social life within the community. In a qualitative research study exploring the needs of parents who are working full time and caring for a child with chronic illness, there is a direct negative impact on parental work life (George, Vickers, Wilkes, & Barton, 2008). The study revealed that parents had to rearrange their work schedules, work at alternative hours, decrease their work hours, change their jobs and even sacrifice their careers due to their attempts to balance their various roles (George, Vickers, Wilkes, & Barton, 2008).

Research on families of children diagnosed with a chronic illness suggest caregivers suffer a significant burden as result of the care, the increasing demands and restructuring of roles that the illness may require (Compas, Jaser, Dunn, & Rodriguez, 2012). Caregiver burden in the context of chronically ill children is a result of the psychological and social effects that a diagnosis has on the individual providing the care to the child and the rest of the family (Buhse, 2008). These effects occur at the onset, at diagnosis and during the course of the disease causing stress, worry, grief, anxiety, and possible depression (Buhse, 2008). As the disease progresses, worry concerning

irreversible, physical, and psychological symptoms as well as latent side effects from medications and treatment becomes apparent. Chronic sorrow and fear of uncertainty begins to take precedence in the lives of caregivers (Gannoni & Shute, 2009; Hobdell, 2004; Wollenhaupt, Rodgers, & Sawin, 2012).

After the diagnosis of a chronic illness, life for the child and family begins to settle and families begin the transition to living with the diagnosis of a chronic illness. The initial experiences are unsettling to the family because the experiences are new and unexpected. The families make physical, social, financial, psychological and emotional adjustments to the diagnosis (Compas et al., 2012; Goldberg & Rickler, 2011). Families begin to adjust and find ways to cope with the diagnosis of a chronic illness and decrease the amount of stress it has on the family (Compas, et al., 2012; Lawrence, 2012). Families become educated about the symptoms of the disease and learn how to deal with these symptoms. They utilize resources previously in place as well as additional resources so that they can adjust to the situation that the disease may impose. Families develop the skills and capabilities to provide optimal care to the child with the chronic illness while establishing a sense of normalcy in their family life (Lawrence, 2012). The long-term goal in the chronic illness is for families to move toward successful adaptation and the development of family resilience (Black & Lobo, 2008). Family resilience is the successful coping of family members as a unit under adversity that helps them to survive and exist by supporting and comforting each other while remaining cohesive (Black & Lobo, 2008).

Problem and Significance

Multiple sclerosis (MS) is a chronic autoimmune disorder of the central nervous system causing demyelination, inflammation and progressive debilitation if it is not treated with disease altering medications (Boyd & MacMillan, 2005). It predominantly effects Caucasian females between the ages of 20 and 40 years of age (Eckstein & Bhatti, 2016; Ross, Halper & Harris, 2012). Over the last two decades, there have been documented cases of MS affecting other groups of individuals including those under the age of 18. It is estimated that 2-5% of the entire United States (US) population (N= 400, 000) of individuals afflicted with MS have pediatric onset multiple sclerosis (POMS) (Narula & Banwell, 2015). Most children, adolescents and teens with POMS have a relapsing course of the disease (Chitnis, 2013a). The course of the disease is characterized by periods of attack or exacerbation (relapse) followed by periods of complete or partial recovery (remission); hence it is called relapsing remitting MS (Chitnis, 2013a). Because the onset of a relapse is unpredictable, cognitive and physical disabilities can occur unexpectedly, leading to extra outpatient visits and for some, hospital admissions (Boyd & MacMillan, 2005). The severity of the disease varies for each individual based on his or her immune response and the nature of the relapse. The child or teen has to be evaluated and treated by health professionals for the relapse, which results in school absences and disrupted academic performance (Boyd & MacMillan, 2005).

Furthermore, because of the physical disabilities that the child may experience such as imbalance, lower extremity weakness, issues with dexterity, and visual disturbance, this will hinder the child's ability to provide self-care is impacted and

normal social activities are disrupted. These factors could lead to poor quality of life, lower self-esteem, and loss of independence because the length of time of a relapse can be from weeks to months with symptoms that may be temporary or permanent with residual effects (Boyd & MacMillan, 2005).

As with most chronic childhood illness, POMS not only affects the individual child, adolescent and teen but can also cause temporary and/or permanent disruption in the structure and function of the family (Boyd & MacMillan, 2005; Buhse, 2008; MacAllister, Boyd, Holland, Milazzo, Krupp, et al., 2007). Existing literature identifies the physical and psychological effects of POMS on the child, but the effects on the family has yet to be explored. One phenomenological study focused on capturing the lived experience of children living with POMS acknowledged that children with POMS are even concerned with parental involvement in their on-going care (Boyd & MacMillan, 2005). However, the study does not indicate any specific concerns nor does it indicate the extensiveness of parental or caregiver involvement with a child with POMS. Because of the unique characteristics of POMS and the unpredictability of the disease, the experiences of the families of children with POMS may differ from the experiences of other families. Little if anything is known as this phenomenon has not been explored in the literature. The initial and ongoing psychosocial, financial, emotional and behavioral impact on families has not been explored as it pertains to the family of children with POMS. Firsthand accounts of families living the experiences should be examined to explore a disease of this nature and its effects on the function and structure of the family and to discover the influences of the families to adapt or maladapt to the demands of an unpredictable disease. Furthermore, the social and psychological adjustments that

families have make to cope and balance the needs of the child and the rest of the family have not been explored either. To determine the adaptive capabilities that the families utilize to become resilient after a diagnosis of POMS, strategies and resources that the families of children with POMS should be explored as well.

Caregiver's Perspective

There are many definitions of a caregiver. The Oxford English Dictionary defines caregiving as the character of attention to the needs of others, especially those unable to look after themselves adequately ("Caregiving", 2010). In the case of children with chronic illness, the caregiver's perspective of a child with a chronic illness is a vital component to understanding the child and family's initial and ongoing experiences with a diagnosis (Goldberg & Rickler, 2011). The caregiver provides one-on-one care and can witness the child's physical and psychological response to the manifestations of the disease, but also can provide accounts of the response of others in the household as it pertains to the family. In addition, caregivers can also offer insight on family relationships, social support, family resources, stressors, communication and coping, which all may influence how well a family adapts to and copes with a diagnosis (Goldberg & Rickler, 2011). The caregiver establishes rapport with the healthcare providers and keeps the other family members informed of the treatment regimen that deemed best for the child with the chronic illness (Goldberg & Rickler, 2011). The caregiver is usually the person who discusses with the other adults the routine for the family and offers insight into the adjusted role of the family members (Goldberg &

Rickler, 2011). A primary caregiver can take on many forms. The caregiver may or may not be biologically related to the child and may not be a parent or stepparent.

Study Purpose

The purpose of this qualitative descriptive study is to explore caregiver perspectives of how family factors influence adaptation in families of children with pediatric onset multiple sclerosis (POMS) seen at a southeastern specialty center. Family factors are those factors that shape the family process and outcomes of adaptation (McCubbin, Thompson & McCubbin, 2001). Over the years, family research and conceptual literature on family adaptation and resilience have identified the following factors that influence adaptive processes in the face of adversity and illness: family demands or stressors, family types, family resources, appraisal and family problem-solving, communication and coping, financial management, routines and rituals and social support (Black & Lobo, 2008). This study will identify which perceived factors are relative to families of children with POMS.

Specific Aims

The specific aims of the proposed study from the caregiver's perspective are to:

1. Explore the initial and ongoing psychosocial, financial, emotional and behavioral impact on families who have a child with POMS;
2. Explore how the family adjusts to raising a child with POMS;
3. Explore strategies that are utilized for coping and balancing the needs of the child with POMS and their family; and

4. Identify the resources that families perceive are available to families of a child with POMS.

Central Research Questions and Sub-questions

The research study will be guided by the following central research question: How do family factors influence adaptation in families of children diagnosed with multiple sclerosis?

The central question will be further supported by the following research sub-questions:

1. What are the stressors and strains experienced by families of children with POMS?
2. What are the strengths and capabilities of individual family members, the family working as a unit and within the community?
3. How do caregivers of children with POMS appraise or view the experience of raising a child with POMS?
4. What are caregivers' perceptions of their ability to manage the demands associated with the medical management of a child with POMS?
5. What strategies does the family use to cope with the ongoing challenges associate with raising a child with POMS?
6. Regarding the immediate family members, how well are families of children with POMS able to achieve a balance between the needs of the child with POMS, the needs of the family as a whole and the needs of other family members within the home?

7. How do caregivers describe the availability of resources to assist families' adaptation to raising a child with POMS?

Theoretical Framework

The Resiliency Model of Family Stress, Adjustment and Adaptation (The Resiliency Model) will be the theoretical framework that will be used guide the development of interview questions to explore adjustments, adaptations and maladaptation in families of children with POMS. The Resiliency Model was developed in 1989 as a measure of family dynamics related to studies of family resilience and stresses in various setting as it pertains to chronic illness in children and families of children (McCubbin et al., 2001). The model was the result of inductive theory building in which there has been continuous efforts to introduce, revise and develop through hypothesis testing that lead to major sources of concepts and propositions contributing to the advancement of this theory (McCubbin & Patterson, 1983). There are two conceptual phases of the model. The first phase is the adjustment phase in which families attempt to maintain patterns that guide their day-to-day activity (Cardoso & Chronister, 2009). The second conceptual phase, the adaptation phase, involves the families' attempts to maintain the patterns of function that occur over time (Cardoso & Chronister, 2009). Both phases will be examined in the proposed study and factors such as family demands, stressors, resources, appraisal, problem solving skills and methods of coping will be examined to understand how families achieve balance and adapt to a diagnosis.

Design and Methodology

To answer the proposed research question, a qualitative descriptive research study will be conducted through purposeful sampling of primary family caregivers of children who have been diagnosed with POMS and seen at the Center for Pediatric Onset Demyelinating Disease (CPODD) or at the University of Alabama (UAB) Adult Multiple Sclerosis Center, both of which are primary sites of recruitment. The method of inquiry will be one-on-one, in-depth semi-structured interviews with the caregivers of children with POMS. In addition to the interview, the caregivers will complete a 50-item socio-demographic form.

Qualitative inquiry will allow for a rich and in-depth account of the factors that influence that adaptation or in some cases maladaptation of families of children with POMS, which is a phenomenon that has never been explored before (Sandelowski, 2000). This method will be an effective way to bring meaning and understanding to the families' experiences and to have a firsthand account of their encounters that are experienced over time. Qualitative descriptive inquiry will allow the researcher to identify common themes among the families of the children with POMS that will provide the foundation for further inquiry or lead to the creation of family interventions that will increase effective coping and positive family adaptation in the presence of POMS.

Summary

Chapter 1 provided an overview of the problem with support of the importance and the impact that it has on families of children with POMS. This chapter also introduced the problem and the proposed study. A brief overview of the theoretical

framework was also presented that will guide the researcher's interview questions and the research design with rationale for the chosen method. Chapter 2 will detail a review of relevant literature as it pertains to this study and will provide detail regarding the conceptual framework that will be used to guide various aspects of the study.

CHAPTER 2

REVIEW OF LITERATURE

To understand the nature of the literature reviewed in this chapter, one must first understand the epidemiology of MS. This chapter provides the reader with an overview of multiple sclerosis as it pertains to all persons afflicted by the disease. The overview explores the following: history of MS, etiology, disease course, clinical manifestations, diagnostic criteria, and currently available treatment options. This overview will provide the reader with a better understanding of the adaptations that may be necessary after a diagnosis of MS is made. In addition, this chapter will provide the theoretical framework that will support the underpinnings for the proposed study. The overview of the theoretical framework will give the reader insight into how the author will shape the review of literature and the research questions. Furthermore, the author will provide an integrative literature review of the broad and general context of factors that influence the adaptations of families of people living with MS and provide insight on gaps in the literature that will be addressed with the proposed study. The review of literature on the adaptation of individuals living with MS and their families will lead to the identification of gaps in the literature regarding the rare and understudied disease of POMS. Finally, the chapter will provide insight to the reader about the adaptability of families of a child diagnosed with juvenile idiopathic arthritis (JIA), an autoimmune disease that typically affects adults but takes a very similar pattern of progression in children to that of POMS.

Overview of Multiple Sclerosis

History of Multiple Sclerosis

Jean Marie Charcot, a French neurologist, documented the first case of multiple sclerosis in adults in 1848. Although this was the first case to be documented in writing, Jean Cruveilhier, Charcot's mentor later revealed that he was aware of an undocumented case in 1829 (Landtblom, Fazio, Fredrikson, & Granieri, 2010). Charcot identified a triad of symptoms (nystagmus, intentional tremor and aphasic speech) in one of his maids and these symptoms remain among the list of symptoms that patients with multiple sclerosis may experience today.

The first cases of MS in children were confirmed by autopsy and reported in the late 19th century (Hanefeld, 2007). Pediatric MS was often dismissed as a diagnosis in living children due to the difficulty in differentiating among leukodystrophies, inherited metabolic disorders, infectious and post-infectious diseases, and other childhood illnesses that mimicked the symptoms of MS. As a result, many cases of POMS were misdiagnosed until later proven by autopsy. The emergence of magnetic resonance imaging (MRI) technology and subsequent development of diagnostic criteria for adult MS has increased recognition that MS can begin in childhood (Narula & Banwell, 2015; Przybek, Gniatkowska, Mirowska-Guzel, & Członkowska, 2015). Based on an estimated prevalence of 2-5% of MS beginning before age 18, there may be as many as 5,000 - 10,000 cases of pediatric onset multiple sclerosis in the United States alone (Belman, Chitnis, Renoux, & Waubant, 2007).

Etiology of Multiple Sclerosis

Multiple sclerosis is believed to be an autoimmune disorder but has an unknown etiology. It is thought to be the result of an interaction between environmental triggers and predetermined genetic factors that cause the disease to manifest in an individual (Tullman, 2013). The interaction leads to an immune response that results in the destruction of the covering around nerve fibers (myelin sheath) as well as neurons, axons and oligodendrocytes (Tullman, 2013). Over the years, environmental risk factors, such as infectious and viral agent exposure prior to the onset of puberty, sunlight exposure, and low vitamin D levels, have been the topic of epidemiological research. Infectious and viral agents such as herpesvirus 6, Epstein Barr virus and mycoplasma pneumoniae as well upper respiratory track and bacterial urinary tract infections have been suggested as potential triggers (Tullman, 2013). Although MS is not a hereditary disease, there is an increased risk of 1.2-2.0% fold associated with having a first degree relative (child, parent or sibling) with the disease (Tullman, 2013). Because MS is a multifaceted and complex disease, ongoing epidemiological research continues to explore potential etiology in hopes of establishing innovative treatments and even a cure.

Disease Course

There are four types of multiple sclerosis and each type has a distinctive disease course. The most common type, affecting 85% of adult MS patients, is relapsing remitting multiple sclerosis (RRMS) (Dutta & Trapp, 2014). This course is biphasic in nature with alternating periods of acute attack (also known as relapse) followed by partial or complete recovery. An acute attack or a relapse is when an unexpected new symptom appears or the worsening of an existing symptom occurs and last 24 hours or more (Ross

et al., 2012). Complete recovery occurs when a person suffering from their attach makes a full recovery within 6 months with no residual deficits (Ross et al., 2012). Partial recovery means that the person suffering from the symptom(s) did not return back to their baseline exam within a reasonable amount of time of at least six months and has some residual deficits that can be observed on examination (Ross et al., 2012). In 60% to 70% of adults, the relapsing remitting course transforms into a secondary progressive course of the disease (SPMS) after twenty years or more. (Dutta & Trapp, 2014). With this course, there is a progression of symptoms and a decrease in recovery even in the absence of relapse (Dutta & Trapp, 2014). The other forms of MS are primary-progressive MS (PPMS) and progressive-relapsing MS (PRMS). PPMS, which effects 10% of the adult MS population, is characterized by a progressive neurologic dysfunction from the beginning stages of the disease without distinct relapses or remission (Dutta & Trapp, 2014). PRMS, which is the least common of the four courses, is characterized by steady and progressive disease from the beginning with occasional exacerbations along with recovery; however as this course progresses, stages of remission cease (Dutta & Trapp, 2014).

Ninety-five to 99% of the patients with POMS have RRMS. Longitudinal research on POMS suggests that pediatric patients have an annualized relapse at a rate of 2 to 3 times higher than adult patients during the first two to three years of their disease because POMS is more inflammatory in nature (Benson et al., 2014; Chitnis, 2013a). There are a few documented cases of SPMS in pediatric patients, representing less than 1% of the POMS population (Beres, Graves & Waubant, 2014). Research suggests that the age at which actual disability progresses is ten years earlier in the POMS population

than in those individuals who develop MS in adulthood (Chitnis, 2013a; Renoux et al., 2007). Although, there are no documented cases of PPMS or PRMS in children in the United States in the literature, there are documented cases in Iran and Iraq (Etemadifar, Afzali, Tabrizi, & Hosseini, 2013). It is not known if these cases are related to geographic location or ethnicity, as studies of this nature have not been conducted.

Other interests to researchers and health professionals who care for individuals with MS, is the concept of benign MS. Patients with benign MS have very rare attacks and remain minimally disabled for at least 20 years following diagnosis (Hutchinson, 2012).

Clinical Manifestations of MS

MS can affect white and grey brain matter of the central nervous system (CNS) including the spinal cord, optic nerves and brain. An attack may be localized to a single region of the CNS (mono-regional) although demyelination of densely innervated regions such as brainstem or spinal cord may cause widespread impairments. Poly-regional relapses are caused by attacks involving multiple regions of the CNS at the same time. A small subset of POMS presents with acute disseminating encephalomyelitis (ADEM) initially, in which the child has severe encephalopathy and poly-regional deficits.

The most common symptoms of MS include cognitive impairment, sensory dysfunction, alterations in mood, sleep disturbance, hearing impairment, visual loss, movement disorders of the eye, optic neuritis, diplopia, motor weakness, spasticity, ataxia, balance issues, paresthesia, sexual dysfunction, and bowel and bladder dysfunction (MacAllister et al., 2007). The more common but less difficult to localize symptoms include fatigue, depression, pain, heat, and cold sensitivity. Less common

symptoms include impaired speech, swallowing and gag problems, tremors, seizures, breathing problems, itching, headache and hearing loss. Symptoms of MS may be temporary or permanent, and vary based on the severity of each relapse and the individual patient's immune response (Tullman, 2013). Symptoms are variable from person to person. An individual with MS may experience only one or two of the possible symptoms while others experience more symptoms (MacAllister et al., 2007). Adults with MS tend to experience more physical symptoms with their relapses. On the other hand, children with POMS tend to experience more of the "silent symptoms" such as fatigue, major depression and cognitive dysfunction (MacAllister et al., 2007).

Diagnostic Criteria for MS

Because of the multi-faceted nature of MS, researchers developed criteria for diagnosing MS to standardize the diagnosis (Mattson, 2002). The first official criteria for the diagnosis of MS were developed by the Schumacher Committee in 1965 and were purely based on clinical findings. These clinical findings had to be documented as being "separated by space and time". This meant that more than one documented clinical event occurred in at least two separate areas of the central nervous system at least one month apart from each other in the clinical history (Mattson, 2002). During the next three decades, scientific discoveries led to the knowledge that lesions on the brain and spinal cord may cause asymptomatic damage (Mattson, 2002). As technology advanced, nerve evoked potentials and spinal fluid evaluation were used to document this damage and were used along with the clinical findings to confirm the diagnosis (Mattson, 2002). For the first time, capturing silent symptoms lead to diagnosing individuals with possible, probable and definite MS (Mattson, 2002). In 2001, with the advances in radiologic

imaging, the McDonald Diagnostic criteria were developed. The criteria included clinical symptoms along with MRI findings and the exclusion of other disorders that may mimic MS (Mattson, 2002). The McDonald Diagnostic criteria were revised in 2005 and 2010. In 2013, the International Pediatric MS Study Group updated the criteria for pediatric multiple sclerosis based on the 2010 McDonald criteria (Chitnis, 2013a).

Multiple sclerosis is considered a diagnosis of exclusion because most of the symptoms of MS are common to other disorders (Toledano, Weinshenker, & Solomon, 2015). The list of differential diagnoses for MS include but are not limited to infections or inflammatory disorders of the central nervous system (CNS), leukodystrophies, vasculopathies, metabolic disorders, rheumatological disorders, genetic disorders, brain tumors, elemental deficiencies, structural damage in the brain and spinal cord, and other demyelinating disorders of the CNS (Toledano et al., 2015). The differential diagnosis of MS in children and adolescents mimics a larger number of disorders as well, which can lead to a misdiagnosis or delayed diagnosis of MS (Hahn, Pohl, Rensel, & Rao, 2007). As a result, extensive laboratory testing and serial radiographic follow-up may be indicated prior to a definitive diagnosis of MS (Hahn et al., 2007).

Current Treatment Options

There is currently no cure for any variety of MS (Gohil, 2015). The goal of medications used in the treatment of multiple sclerosis is to reduce the number of relapses, slow disease progression, and delay the onset of disability (Derwenskus, 2011). Although documented cases of this disease occurred over a century and a half earlier, it was not until 1993 when the US Food and Drug Administration (FDA) approved the first subcutaneous injectable medication, interferon beta-1b, for adults. This medication alters

the course of the disease, thus it was termed a disease modifying therapy (DMT) (Eckstein & Bhatti, 2016) . Prior to this time, individuals with MS were crippled by the progressive and unpredictable nature of the disease due to repeated attacks on the neurological system. In the mid-1990's, three additional injectable DMTs (interferon beta-1a, intramuscular and subcutaneous) and glatiramer acetate, were approved for adults with MS and became the first line therapy for RRMS (Eckstein & Bhatti, 2016). In addition, five other injectable DMTs were approved for the treatment of RRMS (Eckstein & Bhatti, 2016). These medications were either a new formulation of the original first line therapies or were used to treat RRMS if an individual was unresponsive to two or more of the other therapies (glatiramer acetate, three times per week), generic interferon beta-1b, pegylated interferon beta-1a, generic daily glatiramer acetate and daclizumab (Eckstein & Bhatti, 2016). The discovery of genes that link MS to T-cell and B cell mediated responses lead to the discovery of two intravenous medications, mitoxantrone and alemtuzamab. These two medications were approved in the US by the FDA for second line treatment in adults with relapsing remitting MS who had worsening RRMS or who inadequately responded to at least two other DMTs (Eckstein & Bhatti, 2016). Natalizumab, a third intravenous medication that works to interfere with leukocyte migration across the blood brain barrier, was FDA approved in 2006 (Eckstein & Bhatti, 2016).

In 2010, the first oral agent, fingolimod was approved for the treatment of RRMS in adults (Eckstein & Bhatti, 2016). Since then, two other oral agents, dimethyl fumarate (2012) and teriflunomide (2013) became FDA approved (Eckstein & Bhatti, 2016). In 2017, ocrelizumab, an intravenous agent indicated for the treatment of relapsing remitting

multiple sclerosis as well as for the treatment of primary progressive MS to be given every six months, was approved totaling 15 FDA approved medications for the treatment of RRMS (Eckstein & Bhatti, 2016; Hauser et al., 2017; Wingerchuk & Weinshenker, 2016). Prior to 2017, there were not medications indicated for the treatment of any form of progressive MS (Montalban et al., 2017). The brand name, route, and frequencies of the 15 currently approved DMTs are listed in Table 1.

Treatment side effects are variable and range from headaches, increased risk of infection, transient elevated liver enzymes, diarrhea, pain and skin reaction at the injection site, hair thinning, flushing of the skin, nausea, vomiting, diarrhea and flu-like symptoms to serious but rare side effects such as a brain infection or the risk of human fetal damage (Eckstein & Bhatti, 2016). In addition, patients may develop antibodies against these medications that eventually reduce the efficacy of the therapy and increase the number of relapses leading to disease progression (Strayer & Carter, 2012). Because of these potential side effects, laboratory follow-up is necessary to determine if there is a need to change therapies (Eckstein & Bhatti, 2016). Complete blood count, liver function testing and interferon beta neutralizing antibody testing are routine blood tests that are obtained to assess for elevated liver enzymes, low white blood cell counts or antibodies that have developed that reduce the effectiveness of interferon therapy (Moses & Brandes, 2008).

In addition to laboratory testing, frequent routine history and physical examination as well as serial MRIs are obtained assess for active disease and thus the need to transition to another therapy or to advance to a second line therapy (Narula & Banwell, 2015). Individuals, families, caregivers, employers and the entire healthcare

system carry a substantial economic burden associated with this disease over a period of many years (Owens, 2016). The cost of the first line medications is estimated at $60,000 per year prior to out-of-pocket expenses for the families of persons living with MS (Hartung, Bourdette, Ahmed, & Whitham, 2015).

Although not FDA approved for use in children and adolescents, first line therapies are frequently used off-label in the US and in Europe because of long standing data from adult clinical trials proving the safety and efficacy of these medications (Narula & Banwell, 2015). The therapies are subcutaneously or intramuscularly administered injections. This along with the fact that they are administered in frequencies ranging from daily to weekly can be particularly challenging in the pediatric population. The potential for anxiety due to needle phobia and the possible side effects from these medications can evoke anxiety and uneasiness for the child and family. For these families, medication costs may be associated with high co-payment and deductibles as well as lengthy prior authorizations and pre-approvals based on insurance coverage since none of the medications are indicated in children. Recent legislation in the US has mandated clinical studies for all new therapeutics applicable to children and several clinical trials in children are underway that will provide valuable information regarding the safety and efficacy of newer drugs (Chitnis et al., 2016).

Second line therapy and oral agents are used with caution in pediatric patients due to the lack of optimal dosing regimens and pharmacokinetic studies in individuals diagnosed with POMS (Chitnis, 2013b). Natalizumab is used rarely in children because of the risk of a rare brain infection, progressive multifocal leukoencephalopathy (PML), that requires extensive monitoring for safety (Simone & Chitnis, 2016). Because of the

heightened immune system of children, they often continue to have breakthrough disease which makes treatment for this population even more challenging (Chitnis, 2013b).

High doses of corticosteroids can hasten recovery from acute attacks bur will not alter the degree of eventual improvement nor prevent further attacks or progressive disability (Narula, 2016; Ross et al., 2012). High dosages of intravenous or oral corticosteroids are given for three to five days, which then may or may not be followed by an oral low-dose steroid taper to prevent steroid withdrawal syndrome (Narula, 2016; Ross et al., 2012). As the steroid doses used for demyelinating relapses are typically 20-30 mg/kg fold higher than doses used for treating other forms of autoimmune exacerbations, high dose steroids are limited to relapses causing functional impairment or physical discomfort (Narula, 2016; Ross et al., 2012). Because POMS patients are at risk for increased relapse rates, they are also at risk of adverse effects from prolonged steroid use including hypertension, diabetes, osteoporosis, and ulcers (Narula, 2016). For children with POMS who do not respond to high dosages of corticosteroids, plasma exchange and/or the administration of intravenous immunoglobulins is initiated (Narula, 2016). In addition to the corticosteroids and the use of first line and second line therapies, other regimens are used to manage symptoms based on the effects of the attack (Samkoff & Goodman, 2011). Physical, occupational and speech therapy may be indicated to treat bladder, bowel or emotional symptoms, fatigue, neuropathic pain, spasticity, depression, tremors, gait disturbance, dizziness, cognitive dysfunction, mood disorders, speech and swallowing difficulties, and ambulation problems (Samkoff & Goodman, 2011).

In summary, an overview of the epidemiology of MS provides a detailed review of MS from a historical and etiological perspective with in-depth information about the

Table 1

Disease Modifying Therapies for RRMS

	Scientific Name	Brand Name & Dosage	Route	Frequency	FDA Approval
First Line Therapies	Interferon beta-1b	Betaseron® 0.25 mg	Subcutaneous	Every other day	1993
	Interferon beta-1a	Avonex® 30 mg	Intramuscular	Once weekly	1996
	Glatiramer acetate	Copaxone® 20 mg	Subcutaneous	Every day	1996
	Interferon beta-1a	Rebif® 44 mcg	Subcutaneous	Three times/week	1998
	Glatiramer acetate	Copaxone® 40 mg	Subcutaneous	Three time/ week	1996
	Generic Interferon Beta -1b	Extavia® 0.25 mg	Subcutaneous	Every other day	2009
	Pegylated Interferon Beta -1a	Plegridy® 125 mcg	Subcutaneous	Every 14 days	2014
	Generic Glatiramer acetate	Glatopa® 20 mg	Subcutaneous	Every day	2015
Second Line Therapies	Daclizumab	Zinbryta® 150mg	Intravenous	Once per month	2016
	Mitoxantrone	Novantrone® 12mg/m^2	Intravenous	Every 3 months with a lifetime maximum of 3 years	2000
	Natalizumab	Tysabri® 300 mg	Intravenous	1 dose every 28 days	2006
	Alemtuzamab	Lemtrada® 12 mg	Intravenous	1 does daily for 5 consecutive days, followed by one dose for three consecutive days one year later	2014
	Ocrelizumab	Ocrevus® 600 mg	Intravenous	Every 6 months	2017
Oral Agents	Fingolimod	Gilenya® 0.5 mg	Oral	Daily	2010
	Teriflunomide	Aubagio® 7mg or 14 mg	Oral	Daily	2013
	Dimethyl Fumerate	Tecfidera® 240 mg	Oral	Twice a day	2013

disease course, clinical manifestations, current diagnostic criteria, current treatment options as well as the economic burden of MS. The overview has provided an understanding of the depth of experiences that families may have deal with, which will set the pace for the need for the review of germane literature on the family factors that exist that may influence how a family copes and adapts to a diagnosis of multiple sclerosis.

Theoretical Underpinning of the Study

This portion of the chapter will include a discussion of the theoretical underpinning supporting the proposed study. The discussion will begin with the origin and history of the basic assumptions of the Resiliency Model of Family Stress, Adjustment, and Adaptation. The concepts relevant to the model will be defined as well as their relationships and how each concept relates to the problem. The section will conclude with a discussion of the rationale for the use of this framework as opposed to other possible frameworks.

The Resiliency Model of Family Stress, Adjustment, and Adaptation

Origin and History

The Resiliency Model is a family level theoretical model developed in 1989 that identifies the assessment of stress and family coping as well as the crisis that has disrupted normal family functioning in the presence of chronic illness in a child or adolescent (McCubbin & McCubbin, 1989). The Resiliency Model stemmed from the original Family Stress Model developed in 1949 by Rueben Hill. Hill's ABCX model of

family stress and adaptation theorized that major stressful events disrupt family equilibrium (Hill, 1949). The ABCX model focused on identifying major contributions to family stress, agents of family stress, and buffers against family stress.

Later family stress models include the Double ABCX Model of Adjustment and Adaptation, the Family Adjustment and Adaptation Response Model (FAAR) and the T-Double ABCX Model of Family Adjustment and Adaptation, which eventually led to the development of the Resiliency Model (Lavee, McCubbin, & Patterson, 1985; McCubbin & Patterson, 1983; McCubbin, Thompson, & McCubbin, 1996; Patterson, 1988). The Double ABCX Model of Adjustment and Adaptation introduced coping behaviors, patterns and strategies into Hill's original family stress theory (McCubbin & Patterson, 1983). Later in the same year, modifications to the Double ABCX model led to the development of the FAAR Model, which integrated coping, incorporated the consolidation phase in the previous stress model and further modified coping strategies into categories of adaptation and adjustment (McCubbin & Patterson, 1983). In addition, the resistance phase and the restructuring phase were introduced into the stress model (McCubbin & Patterson, 1983). The T-Double ABCX Model of Family Adjustment and Adaptation integrated family typologies into the previous model along with additional integration of the life cycle perspective (McCubbin & McCubbin, 1989). The Resiliency Model incorporated relational perspectives of family adjustment and adaptation, family problem solving and family coping and instituted patterns of family functioning as a part of each phase of the model (McCubbin & McCubbin, 1983; McCubbin, McCubbin, Thompson & Thompson, 1995). This framework has been used to study families of children faced with chronic stressors and illnesses such as autism spectrum, cerebral

palsy, Down syndrome, type 1 diabetes, childhood cancer and cystic fibrosis (Brody & Simmons, 2007; Brown, Fouché, & Coetzee, 2010; Greeff, Vansteenwegen, & Gillard, 2012; Hall et al., 2012; Krstic & Oros, 2012; Mitmansgruber et al., 2016). A graphic representation of the model is presented in Appendix A.

Basic Assumptions of the Theory

The Resiliency Model is based on five basic assumptions about family life. The first assumption is that families face hardships and changes that are natural and predictable over the life cycle (McCubbin, Thompson, & McCubbin, 2001, p 14). The second assumption is that families develop patterns of functioning, basic competencies and capabilities that foster the growth and development of its members and the family as a unit in order to protect the family from major disruptions (McCubbin, Thompson, & McCubbin, 2001, p 14). Assumption 3 implies that families develop unique competencies, patterns and capabilities designed to protect family members from unexpected stress or strain (McCubbin, Thompson, & McCubbin, 2001, p 14). This would, in turn, foster the family's ability to recover after a family crisis, which would cause a major transition and change. The fourth assumption states that families have a network of relationships and resources within the community from which they draw during times of stress and crisis (McCubbin, Thompson, & McCubbin, 2001, p 14). The final assumption is that families who are faced with crisis work to restore order, harmony and balance based on the circumstance and the demands for change (McCubbin, Thompson, & McCubbin, 2001, p 14).

The Resiliency Model encompasses two conceptual phases: adjustment and adaptation. During the adjustment phase, families try to maintain patterns that guide their

day-to-day activity such as interaction patterns, rules and roles (Cardoso & Chronister, 2009). The components of this phase that shape outcomes include the following: a) family stressors caused by residual problems in the disabled or ill member b) patterns of family functioning or types and c) vulnerability of families to stress (McCubbin et al., 2001). The individual capabilities of families serve as a buffer to the stress that is imposed on the families by the illness or disability. Capabilities include available resources, coping strategies and positive appraisal. Family adjustment outcomes occur on a continuum with bond adjustment on one end and maladjustment on the other (McCubbin & McCubbin, 1991). Bonadjustment occurs when normal family functioning is maintained and the family has a sense of control over their environment (Cardoso & Chronister, 2009). Maladjustment occurs when the individual family member deteriorates and the family no longer can accomplish life tasks (Cardoso & Chronister, 2009). In the Resiliency Model, family crisis, a direct result of maladjustment, takes place when the family is disorganized after an unexpected situation arises. As a result, there is a demand for changes in family patterns to restore order, coherence and stability, which marks the beginning of the adaptation phase (Cardoso & Chronister, 2009).

The adaptation phase of the Resiliency Model is the concept of understanding the family's struggle to manage the diagnosis of a family member with chronic illness and its effect over time (Cardoso & Chronister, 2009). In this phase, the outcomes of family efforts are to bring a new level of balance, harmony, coherence and an acceptable level of functioning to a family crisis (McCubbin, Thompson, & McCubbin, 1996). The level and capacity of the family to cope in response to a disability or chronic illness onset is based on the following issues: 1) the collective demands of the family, 2) the typology of

the family based on specific strengths, 3) the external resources of the family and 4) the positive appraisal of the situation (Cardoso & Chronister, 2009). Just as with family adjustment, family adaptation occurs along a continuum of outcomes based on the family's determination to find balance (Cardoso & Chronister, 2009).

Coping, refers to the specific cognitive and behavioral efforts in which the individual and family attempt to reduce or mange the demands placed upon them (Mu, 2005). Coping strategies used by families can serve as an indicator of adaptation to a disease process (Krstic & Oros, 2012). According to the Resiliency Model, coping strategies involve two levels of interaction: 1) between the individual and the family, which is the internal strategy, and 2) between the family and social environment, which is the external strategy (Krstic & Oros, 2012).

The adjustment phase of the Resiliency model demonstrates how the family responds to events that may not cause major hardships or may be the initial response of the family to a major event that has taken place. The adjustment phase begins with the onset of an illness or stressor that affects the family of an affected child. In this phase, families attempt to maintain patterns that guide day-to-day activity that define interaction, rules, and roles (Cardoso & Chronister, 2009). This phase consists of the variables of the stressor, vulnerability, established patterns of functioning, family resources, family appraisal of the stressor, problem solving and coping, and the outcome of adjustment. All these components determine if the outcome is bonadjustment (balance and harmony) or maladjustment (imbalance and disharmony) (McCubbin et al., 2001).

On the other hand, the adaptation phase focuses on how a family responds to a serious event that will require major transition or hardship and necessitate change to

occur systematically over time. The process of adaptation in the presence of chronic illness includes the process of change that occurs in the environment, the community and the family's relationships to the community to restore harmony, balance and well-being within the family (McCubbin et al., 2001).

All components of the adaptation phase (patterns of functioning, levels of appraisal, resources, social support, coping and problem solving) interact with each other and shape the level of adaptation. The adaptation phase begins with an introduction of a stressor into the family. The family responds based on their appraisal of the stressor and the resources available to the family to problem solve and cope with the stressor. Initially, the families may maladjust to the crisis based on the pile up of demands, inadequate patterns of family functioning or a deterioration in a previously adequate level of functioning. Over time, families either retain, restore or develop and institute new patterns of family functioning. The result is either bondadaptation, which indicates positive coping and adaptation, or maladaptation, which indicates that there is a continued crisis and a need for assistance or referral for help with the crisis. For the aim of the study, the adjustment and adaptation phase of the Resiliency Model are applicable to families of children with POMS because families' experiences are different and are based on the various stages in the disease process.

Definition of Concepts Related to the Theory

According to the Resiliency Model, two families experiencing the same situation or crisis may respond differently based on several factors that may influence the process of adjustment and adaptation. These factors include *family demands or stressors, family*

typology, family resources, family appraisal and family problem-solving communication and family coping. The success of the families' adaptation is based on these factors and will be discussed. In addition, other key concepts such as *vulnerabilities, patterns of functioning, family hardiness, paradigms, schema,* and *pile up of demands* will be discussed as well.

Family Demands

Family demands include the demands on or in the family system due to: (a) a family member having a chronic illness or condition, (b) changes to the family's life cycle (c) any prior unresolved family strains, (d) and consequences of family efforts to cope, (McCubbin, Patterson & Wilson, 1996).

Family Stressor

A stressor is a demand that is placed on the family that either produces or has the potential to produce changes within the family, which may in turn affect all aspects of family life (McCubbin et al., 2001). These aspects include but are not limited to: marital relationships, the parent-child relationships, the sibling relationship(s), the family system boundaries, the goal of the family, and the family's pre-existing patterns of function, balance and harmony. Stressor severity is determined by the degree to which a stressor threatens the stability of the family unit and disrupts the family function, placing demands on the family resources and capabilities (McCubbin, 1986).

Family Type

A family's typology, which is usually a predictable and discernable pattern of family function, is defined as a set of attributes or behaviors that explain how the family behaves (McCubbin, Thompson, & McCubbin, 2001). There are three family types

associated with the Resiliency Theory: the regenerative family type, the resilient family type and the rhythmic family type, all of which have been associated with more adaptive functioning of the family as a unit. The regenerative family type is characterized based on family hardiness and coherence and regenerative families are more likely to display greater domains of bonding, flexibility, family time and routines (McCubbin, Thompson, & McCubbin, 2001). The rhythmic family type is characterized based on family times and routines and the family's value of times and routines and are indicated by satisfaction (marriage, family, child development, family health (mental and physical) and community) and overall well-being (McCubbin, Thompson, & McCubbin, 2001). Resilient family type is characterized based on closeness and flexibility in which they are willing and able to switch roles, and change rules and boundaries as needed (McCubbin, Thompson, & McCubbin, 2001).

Family Resources

Family resources are defined as a family's capabilities and strengths to manage stressors and their demands and to promote balance and harmony to resist crisis while maintaining established patterns of functioning (McCubbin, Thompson, & McCubbin, 2001). The resources include but are not limited to the following: social support, open communication, traditions, celebrations, economic stability, hardiness, flexibility, routine, organization, spiritual beliefs and cohesiveness (McCubbin, Thompson, & McCubbin, 2001). Resources may be on the individual level, family level or community level.

Family Appraisal

Family appraisal is the assessment the family makes of (1) the stressor, (2) the family's capability of managing a crisis, and (3) the family schema's ability to attach

meaning that legitimizes and affirms changes in family functioning (McCubbin, Thompson, & McCubbin, 2001). There are three levels of appraisal in the adaptation phase of the model. The first level of appraisal is the family's appraisal of the stressor, which in the case of chronic illness is the actual medical condition or diagnosis. The second level of appraisal is the family's situational appraisal in which the family makes a shared assessment of the demands, capabilities and the relationship that exists between these demands and capabilities (McCubbin, Thompson, & McCubbin, 2001). Family schema is the third level of appraisal and is defined as a shared set of values, beliefs, rules, goals and priorities that guide and shape the major domains of family functioning. Family schema is more abstract than the other levels of appraisal and reflects how the family appraises a situation based on its collective view of the world.

Family Problem-Solving Communication

Family problem solving is defined as the family's ability to organize stressors into something that is manageable and to identify alternative courses of action to deal with each issue while initiating steps to resolve those discrete issues and interpersonal issues (McCubbin, Thompson, & McCubbin, 2001). This process also occurs while families are developing and cultivating constructive patterns of problem solving and communication that is needed to maintain and restore harmony and balance (McCubbin, Thompson, & McCubbin, 2001). Two types of family problem solving communication are incendiary communication and affirming communication (McCubbin, McCubbin, & Thompson, 1996). Incendiary communication involves screaming and yelling with a lack of calmness and a tendency to bring old issues into the discussion. On the other hand, affirming communication is careful to not cause any physical or emotional harm with

attentive listening in a respectful manner in order to end conflict on a positive note. Affirming communication is the most effective way for families to adapt to situations that may cause stress and strain (McCubbin, Thompson, & McCubbin, 2001).

Family coping is the family's strategies, patterns and behaviors designed to maintain or strengthen the family, maintain the emotional stability and well-being of its members, obtain and use family and community resources to manage crisis situations and initiate efforts to resolve the family hardships (McCubbin & McCubbin, 1993, p. 30). Coping strategies used by families can serve as an indicator of adaptation to a disease process (Krstic & Oros, 2012). According to the Resiliency Model, coping strategies involve two levels of interaction: 1) between the individual and the family, which is the internal strategy and 2) between the family and social environment, which is the external strategy (Krstic & Oros, 2012).

Other Key Concepts

Vulnerabilities are those additional life stressors or changes that may undermine the family's ability to achieve adaptation in the face of family crisis (McCubbin, Thompson, & McCubbin, 2001). *Patterns of functioning* are defined as the elimination, modification, or institution of behaviors by families to arrive at balance and harmony and a satisfactory level of adaptation (McCubbin, Thompson, & McCubbin, 2001). *Pile up of demands* is the accumulation of demands within the family unit such finances, debt, health status of family members, changes in parental work role or environment (McCubbin, Thompson, & McCubbin, 2001). *Family hardiness* is the internal strengths and durability of the family unit that are characterized by a sense of control over the outcome of life events and hardships, a view of change as beneficial and growth

producing and an active rather than passive orientation in responding to stressful situations (McCubbin, Thompson, & McCubbin, 2001). *Family schema* refers to shared values, beliefs, and expectations about family structure, self-/group orientation, spiritual belief, land, nature and time orientation (McCubbin et al., 1995). *Family paradigms* refer to the specific views, expectations, and patterns of functioning affecting specific domains of family life (child rearing, work, education) which may vary by culture (McCubbin et al., 1995).

Relationship between the Concepts in the Theory

McCubbin et al. (2001) identify nine categories of stressors that contribute to pile of demands and vulnerability of a family that contribute to a crisis. These categories determine where or not the family can adapt and adjust to the crisis and ultimately achieve harmony and balance. The categories are as follows:

1. The initial stressor and its related adversities that has developed over time.
2. Normal transitions occurring in the individual or family during the same time period.
3. The accumulation of prior strain
4. Unexpected demands based on the situation and difficulties related to the context of the illness
5. The results of the family efforts to cope that lead to further family burden
6. The inadequate guidelines of the community on how to cope and respond to the crisis as a family

7. The new patterns of family functioning instituted that require change in family functioning
8. The new patterns of family functioning instituted that conflict with the family's values and beliefs and/or rules and expectations
9. Established patterns of functioning that are in conflict or not compatible with recently adopted patterns of functioning

How Concepts Relate to the Problem

Family factors used to adjust and adapt to a diagnosis of POMS have not been explored. The Resiliency Model will guide this study beyond the individual diagnosed with the chronic illness, focusing on risk and resilience of the family as a unit (Walsh, 1996). Using this model will provide awareness and insight as to why some families are more resilient than others are and develop the capability to cope and adapt to the stressors and strains associated with a long term chronic illness such as POMs. The following research questions will be utilized in the study to ensure that the problem is explored based on the model:

Aim 1: Explore the initial and ongoing psychosocial, financial, emotional and behavioral impact on families who have a child with POMS.

- What are the stressors and strains experienced by families of children with POMS?
- What are the strengths and capabilities of individual family members, the family working as a unit and within the community?

Aim 2: Explore how the family adjusts to raising a child with POMS

- How do caregivers of children with POMS appraise or view their experience of raising a child with POMS?
- What are caregiver's perceptions of their ability to manage the demands associated with the medical management of a child with POMS?

Aim 3: Explore caregivers' perceptions of family's strategies for coping and balancing the needs of the child with POMS and of the family.

- What strategies does the family use to cope with the ongoing challenges associated with raising a child with POMS?
- Regarding immediate family members, how well are families of children with POMS able to achieve a balance between the needs of the child with POMS, the needs of the family as a whole and the needs of other family members within the home?

Aim 4: Identify resources that families perceive are available to assist families caring for a child with POMS.

- How do caregivers describe the availability of resources to assist families' adaptation to raising a child with POMS?

The above questions will seek to explore how families adjust and adapt to a POMS diagnosis. This study will seek to identify how the family deals with the pile of demands and stresses specific to their family. It will also identify the family's innate ability to be resilient, how they adapt to change, and how they seek support necessary to cope effectively with the disease as changes occur over time. The proposed study will provide the answers to the questions through the qualitative interview process and the socio-demographic form.

Other Frameworks Considered

Two other models were considered and ultimately rejected to guide the proposed dissertation. The Family Distress Model (FDM) was developed in 1992 as a guide to understand how families go through and accommodate change (Cornille & Boroto, 1992). The model consists of five phases that a family goes through to accommodate change. Phase I is the family's predictable or "stable" patterns that fit their normal and preferable resources, identity, values and goals (Weber, 2011). These patterns are the routines, roles, and rituals that families use for day-to-day decision-making. Phase II is when the normal family patterns of Phase 1 are interrupted and the family experience some level of distress but resolution comes by using preexisting strategies (Weber, 2011). Phase III occurs when a family has a crisis that in turn sends the family into Phase IV where the family withdraws from available social support and becomes intense and preoccupied by the crisis (Weber, 2011). Phase V involves the family seeking social support to cope with the crisis while viewing the disruption that the crisis has caused in the context of the goals of the family (Weber, 2011). During the final phase of the Family Distress Mode, the crisis is resolved or managed and the family can return to their previous pattern in Phase I. Although the Family Distress Model is a family theory model, this model does not address patterns of change that become permanent due to the ongoing challenges of a chronic unpredictable illness such as POMs. For that reason, this theory would not describe the changes that would lead to the adaptation that takes place over time and would not answer the underlying research question based on the family factors that would affect the adaptive process.

The second model considered for this dissertation was the Roy Adaptation Model (RAM). The RAM focuses on adaptation of humans in a continuously changing environment in four adaptive modes (Desanto-Madeya & Fawcett, 2016). The four modes include basic physiological behaviors, self-concept behaviors, role function and interdependence. Basic physiologic behaviors include processes such as nutrition, elimination, protection, and oxygenation (Desanto-Madeya & Fawcett, 2016). Self-concept behaviors are those behaviors associated with physical self (body sensation and body image), personal self (self-consistency, self-ideal) and moral-ethical –spiritual self (Desanto-Madeya & Fawcett, 2016). Role function behaviors are behaviors that are assumed throughout a person's life as well as the activities associated with those roles (Desanto-Madeya & Fawcett, 2016). Interdependence behaviors are those behaviors associated with giving and receiving affection and support in relationships with others (Roy, 2009). In the presence of stimuli, individuals respond within these four modes by integrating, compensating or compromising life processes (Desanto-Madeya & Fawcett, 2016). Adaptation is evident when humans function as a whole in the integrated life process. Although this model deals with adaptation of the individual, it lacks the perspective of the family. This model would not align with the researcher's focus on the family as a unit; chronic illness of children involves the entire family and must be taken in consideration when the researcher aims to capture adaptation and resiliency from a holistic perspective.

Review on Literature on Adults with RRMS and their Families

Search Strategy

The aim of this integrative review is to examine factors that influence adaptation of individuals and their families living with RRMS. To obtain literature about factors related to RRMS, three electronic databases were searched, PubMed, Cumulative Index of Nursing and Allied Health Literature (CINAHL) and Scopus. Key search terms included combining multiple sclerosis (MS) with the following: family coping, family stress, family adjustment, family resources, caregiving, family caregiving, family impact, family adaptation, family demands and family appraisal. The terms were used as keywords and Medical Student Headings (MeSH) to obtain the maximum number of publications. Inclusion criteria were manuscripts reporting empirical research studies (qualitative, quantitative and mixed method studies) that were published in English from January 2006 to August 2016 with a focus on factors in individuals and their families living with RRMS. Reports were excluded if adaptation was discussed in terms of progressive forms of MS (primary progressive, secondary progressive or advanced MS), palliative care, rehabilitation, veteran affairs, long-term care, post-partum, pregnancy, motherhood, end-of-life care, and aging. Reports were also excluded if factors and adaptation to RRMS were discussed in terms of symptoms specific or gender specific issues. Additionally, unpublished manuscripts (abstracts or dissertations) and longitudinal studies spanning more than two years were excluded. Although comparative studies that compare RRMS to other forms of MS or to other chronic illnesses were excluded, studies comparing subjects to healthy participants and those that involved at least half of RRMS participants were examined. Systematic and integrative reviews were

not used as primary data sources but were utilized to obtain additional sources for the review. The CONSORT for this study is outlined in flow diagram in *Appendix B.* Twenty-six studies met the inclusion criteria. A total of 14 qualitative studies, 9 quantitative studies and three mixed methods studies resulted from this review. All the studies involved individuals with MS, their spouse or partner, the children of individuals with MS or the family of as individual with MS as a unit. All the studies that were reviewed individuals with MS that were greater than 18 years of age or their family members, whether it was as a participant or a family member of a participant. The studies were sorted into five categories and were based on the constructs of the theoretical underpinning of the study. The categories included the following: 1) family demands; 2) family resources; 3) family appraisal; 4) family problem solving, communication and coping; and 5) family adaptation. The literature review will be presented based on these constructs. Multiple studies were included within multiple categories as these studies identify more than one construct in the literature.

Factors that Influence the Adjustment and Adaptation of Multiple Sclerosis

This section contains an integrative review of factors (demands, resources, appraisal, problem solving, communication, and coping) that influence the adaptation in families living with the relapsing remitting MS (RRMS). First, the demands that influence an individual's adaptation to RRMS will be discussed. Next, the resources that are available to these individuals and their families from the perspective of family units will be discussed, followed by the appraisal of the disease and then by their problem-solving communication and coping strategies. Adaptation will be discussed last to conclude this review of the literature. The literature review was based on various

perspectives including (a) individuals and caregivers, (b) young caregivers (ages 5 to 18), (c) partners of individuals with MS, (d) family units consisting of the ill parent, the healthy parent and the child, (e) family units consisting of MS patients, their spouses or partners and a child, (f) individuals with MS, (g) dyads consisting of individuals with MS and their significant other, (h) children whose parents were diagnosed with MS and (i) next of kin consisting of spouse or cohabitating partners, parents, siblings and adult children.

Literature Review on Family Demands

When an individual is diagnosed with MS, the family suddenly faces many demands. The demands that MS places on affected individuals as well as their caregivers are well documented in the literature (Aymerich, Guillamon, & Jovell, 2009; Bogosian, Moss-Morris, Yardley, & Dennison, 2009; Fallahi-Khoshknab, Ghafari, Nourozi, & Mohammadi, 2014; Heward, Molineux, & Gough, 2006; Koopman, Benbow, & Vandervoort, 2006; Labiano-Fontcuberta, Mitchell, Moreno-Garcia, & Benito-Leon, 2015; Malcomson, Lowe-Strong, & Dunwoody, 2008; Pakenham & Cox, 2012); however, there is less empirical research with regard to the early phases of RRMS and how it affects the individual, their children and their family as a unit (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016; Diareme et al., 2006; B. Steck et al., 2007). Thirteen empirical studies were included in the literature review regarding the demands associated with RRMS and are presented in Appendix C. Eight of the 13 studies were qualitative, and the remaining five studies were quantitative. Seven of the eight qualitative studies had 8 to 25 participants (Bogosian, Moss-Morris, Yardley, & Dennison, 2009; Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014; Heward et

al., 2006; Malcomson et al., 2008; Turpin, Leech, & Hackenberg, 2008). One of the eight qualitative studies involved 119 individuals with MS and 64 partners, which is atypical of a qualitative study (Pakenham, Tilling, & Cretchley, 2012). The methodology for this study involved an open-ended questionnaire that was sent to 500 individuals with MS and their partners, which yielded the resulting participants. Two of the quantitative studies involved a large number of participants (Aymerich et al., 2009; Koopman et al., 2006). One of those studies involved 705 individuals and 551 caregiver (Aymerich et al., 2009) and the other study involved 353 individuals with MS and 240 significant others (Koopman et al., 2006). The remaining four quantitative studies represented 50 to 150 participants (Diareme et al., 2006; Labiano-Fontcuberta et al., 2015; Pakenham et al., 2012; Steck et al., 2007).

The studies regarding family demands were categorized based on the study participants. Three of the13 studies were conducted with individuals with MS and their significant other or caregiver, usually spouses or partners (Aymerich et al., 2009; Koopman et al., 2006; Pakenham et al., 2012). Two of the 13 studies involved partners of individuals with MS only (Bogosian et al., 2009; Heward et al., 2006) and two involved individuals with MS only (Fallahi-Khoshknab et al., 2014; Malcomson et al., 2008). Three of the 13 studies that were reviewed regarding demands included the individual with MS, their healthy spouse and one randomly selected child, which made up the family unit (Bostrom & Nilsagard, 2016; Diareme et al., 2006; Barbara Steck et al., 2007). Only one study out of the 13 included in this portion of the literature review included an individual with MS, a healthy matched control and a caregiver (Labiano-

Fontcuberta et al., 2015). Two studies out the 13 involved children of individuals with MS (Bjorgvinsdottir & Halldorsdottir, 2014; Turpin et al., 2008).

Analysis of the thirteen studies revealed that the initial demand on adult individuals newly diagnosed with RRMS and their families was due to the shock of the actual diagnosis. In several of the qualitative studies, individuals with MS expressed a sense of helplessness and loss of control (Bogosian et al., 2009; Fallahi-Khoshknab et al., 2014; Koopman et al., 2006; Malcomson et al., 2008). Individuals with MS and families reported having a knowledge deficit that contributed to misconceptions about disease manifestations and clinical outcomes. One study noted that the disease was often concealed because of the misconceptions that led to anger, fear, anxiety and confusion (Fallahi-Khoshknab et al., 2014).

Other stressors that resulted in a strain to the family were: uncertainty of the future, fear of job termination, decreased quality of life, confusion and fear of demoralization and social isolation (Aymerich et al., 2009; Bogosian et al., 2009; Fallahi-Khoshknab et al., 2014; Heward et al., 2006; Koopman et al., 2006). These feelings stemmed from a lack of support and understanding from the family of individuals with MS, friends, and social networks (Bogosian et al., 2009; Malcomson, Lowe-Strong, & Dunwoody, 2008). Aymerich et al. (2009) and Labiano-Fontcuberta et al. (2015), although using different methodologies, both reported that the quality of life of caregivers and individuals with RRMS is lower than before the diagnosis, even in the face of mild disease.

Two of the studies regarding family demands in RRMS identified strategies of the individual and their families that would reduce the stress and strain caused by the initial

and ongoing phases of the diagnosis (Koopman et al., 2006; Malcomson et al., 2008). One of the studies involved 353 individuals with MS and 240 significant others in which a quantitative questionnaire was developed from focus groups and consisted of 75 needs statements (Koopman et al., 2006). The other study was a qualitative study involving 13 individuals with MS who participated in two focus groups to give their personal accounts of their lives with MS (Malcomson et al, 2008). Koopman et al. (2006) and Malcomson et al. (2008) discuss the following needs: 1) psychosocial professional support; 2) unchanging interpersonal relationships; 3) adjustments to current employment circumstances; 4) productive and meaningful life; 5) support of family and friends; and 6) maintenance over control of their life. The families needed reassurance that their MS physician as well as other health professionals, including their primary care provider, were interested in their well-being and that the team did not "feel bothered" by their questions about the disease (Koopman et al., 2006) . The individuals with MS and their families also wanted to receive the knowledge about services that were offered at their MS clinic such as access to support groups, counseling, MS nurses or other specialists (Malcomson et al., 2008).

Two of the six quantitative studies revealed a need to maintain an individual and family social life, which can be difficult after the diagnosis (Koopman et al., 2006; Malcomson et al., 2008). Both studies addressed symptoms such as fatigue or physical disability that hindered individuals with MS and families from maintaining their previous social life. Families oftentimes felt an obligation or loyalty to the person affected by MS and missed events and activities, but the individual with MS felt a need for this social life to be maintained (Malcomson et al., 2008).

Furthermore, individuals with RRMS often felt a need to maintain a productive life and remain financially secure in their current occupation, which encouraged a need for occupational adaptability (Heward et al., 2006; Koopman et al., 2006; Malcomson et al., 2008). According to one study, occupational stability would fulfill an individual's desire to make a contribution to society and aid in that individual's preserved sense of self-worth (Heward et al., 2006). Malcomson et al. (2008) revealed that there might be a need to compromise employment circumstances to balance desire and capability considering an MS diagnosis. Heward et al. (2006) added that partners of individuals with MS identified the need to change their occupation based on the demands and constraints of their loved and the uncertainty of their future.

Not only do the demands of RRMS affect the individual and the family as whole, but also the children of those affected by MS. Six of the eleven articles related to the demands of RRMS addressed the demands that were either placed on children or related to the children within the family of a recently diagnosed individual (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016; Diareme et al., 2006; Pakenham & Cox, 2012; Barbara Steck et al., 2007; Turpin et al., 2008). According to several studies (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016), children were impacted by the symptoms that their parent with MS experience. The acute and unexplained symptoms in parents with MS created anxiety in their children (Bostrom & Nilsagard, 2016). The children faced increased demands to provide care for their parent and this was often embarrassing and difficult for both the parent with MS and the child due to the their age (Bjorgvinsdottir & Halldorsdottir, 2014). The children were also concerned about their parent and their families' future and worried about parental death.

Lack of knowledge about the disease resulted in misunderstanding, which, in turn, led to emotional issues such as anxiety and depression (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016; Diareme et al., 2006; Steck et al., 2007; Turpin et al., 2008). Diareme et al. (2006), using a quantitative comparative methodology with 56 family units consisting of individuals with MS, their spouses and one randomly selected child (ages 4 to 17), demonstrated that the negative effects associated with family dysfunction and problems that children of individuals with MS experience are more frequent in poorly adjusted than well-adjusted families.

After the initial phase of the diagnosis passed, as adjustments to the diagnosis are made, children of individuals with MS experienced a wide range of emotions that were dependent on their developmental level (Bostrom & Nilsagard, 2016; Pakenham & Cox, 2012). Some older children became engaged and interested in assisting their parents and family while the younger children became angry and obstinate because of the unexpected changes (Bostrom & Nilsagard, 2016; Pakenham & Cox, 2012). Because of the shift in parental or family responsibilities, parental attachment issues arose because the younger children did not understand the interruption in their relationships. Two studies discussed parental guilt and personal neglect of self in individuals with MS (Bostrom & Nilsagard, 2016; Pakenham & Cox, 2012). In addition, individuals with MS with children had initial concerns unrelated to themselves and their personal health status, but rather to the needs of their children (Bostrom & Nilsagard, 2016; Pakenham & Cox, 2012). This included the need to address the reaction of their children to the diagnosis and how the children may adapt to their parents' diagnosis and management of MS (Bostrom & Nilsagard, 2016; Pakenham & Cox, 2012). There were also concerns about children's thoughts

about their parent and family future (Bostrom & Nilsagard, 2016; Pakenham & Cox, 2012).

Teenagers of individuals with RRMS may experience greater emotional and behavioral problems than their peers. Three of the studies on demands in this review suggested that teenagers take on additional roles and responsibilities, which restrict their participation in developmentally appropriate activities (Bostrom & Nilsagard, 2016; Diareme et al., 2006; Turpin et al., 2008). The roles included activities such as an increase in household chores or greater responsibility with their siblings. This may be distressing to both the teen and the parent with MS as well as other family members because outward displays of distress such as sorrow, depression, grief, tension, and anger may be manifested. Bjorgvinsdottir and Halldorsdottir (2014) suggested that teenagers of individuals living with MS who become primary caregivers feel uninformed and isolated and, oftentimes, unsupported at school and home.

The thirteen studies discussed have provided evidence on the psychological, physical and social demands that are placed on the family when a member of the family is diagnosed with MS. Although the Resiliency Model is not cited as a guide in any of the studies, all the studies in this review support the fact that family demands due to the physical and psychological manifestations are stressors that may inhibit a family from adjusting to the diagnosis of MS. From the time of diagnosis toward the ongoing phases of the disease, families are faced with feelings of helplessness, loss of control, anxiety, confusion, decreased quality of life, and social isolation, which are all outwards signs of stress and strain on the family. Shifts in roles and responsibilities of all family members and the increase in unexpected and unmet needs (i.e. professional support, stable

interpersonal relationships, employment adjustments, family support, and social life) may cause families to maladapt if adjustments are not made to the family environment.

Literature Review on Family Resources

Individuals with MS and their families have resources that give them the strength and capability to better manage the demands and stress of the diagnosis (Diareme et al., 2006; Eliášová, Majerníková, Hudáková, & Kaščáková, 2015; Ghafari, Khoshknab, Norouzi, & Mohamadi, 2014; Horton, MacDonald, Erickson, & Dionigi, 2015; Mulligan, Wilkinson, & Snowdon, 2016; Pakenham et al., 2012). Although these strengths can be on the individual level, more research has been conducted on the interpersonal and community level. Six empirical studies were included in the literature regarding resources in the adjustment and adaptation of individuals with RRMS and their families and are presented in the table in Appendix D. Four of the six studies were qualitative in nature and the remaining two studies were quantitative. Two of the six studies were previously mentioned in the discussion of the literature regarding family demands (Diareme et al., 2006; Pakenham et al., 2012). Of the four studies that were not mentioned in the review on family demands, one involved the review of resources as it pertains to individuals with MS and their spouses (Horton et al., 2015) and three of the studies were with regard to individuals with MS (Eliášová et al., 2015; Ghafari et al., 2014; Mulligan et al., 2016).

Three of the six studies identified a list of resources that can be utilized to balance the new demands faced by individuals with RRMS and their families (Diareme et al., 2006; Pakenham & Cox, 2012). Assistance from friends and family with chores, physical support, physical care, financial support and time management, as well as practical assistance (i.e. help with budgeting, job search or employment of a housekeeper) were

examples of a few resources mentioned (Ghafari et al., 2014; Pakenham & Cox, 2012). Specific assistance from spouses offered a resource for couples and a sense of hope. Considering children of individuals with MS, positive family functioning was identified as a protective factor in the development of aggressive and delinquent conduct in children of individuals with MS (Diareme et al., 2006).

The other three studies reviewed in the literature on resources are intervention studies (Eliášová et al., 2015; Horton et al., 2015; Mulligan et al., 2016). One of the intervention studies evaluated the effectiveness of individuals with MS who attended a self-help group as compared to individuals who did not attend a self-help group as the control group control group (Eliášová et al., 2015). This study revealed that the group of participants who received the self-help intervention had improved scores in the their quality of life in three domains (physical health, survival and social relationships) (Eliášová et al., 2015). Self-help groups along with the support of family and health care professionals allowed individuals with MS to lead nearly normal lives. The other two interventions studies were self-management programs regarding fatigue and physical impairment, two of the most common symptoms experienced by individuals with MS (Horton et al., 2015; Mulligan et al., 2016). The fatigue self-management program, when integrated in the daily lives of individuals with MS, had positive effects on the lives of the participants as well as their families as determined by the two themes that emerged from the analysis of the study: the achievement of behavior change to manage fatigue and the "whole of life effects" (Mulligan et al., 2016). The participants achieved behavior change to manage fatigue by reflective learning, taking control and developing new habits, while achieving "whole life effects" by building resilience, obtaining balance in

their lives, improving family dynamics and a new support network (Mulligan et al., 2016). In the exercise intervention study, regular physical exercise intervention twice per week within a group as well as following their own individualized program improved both physical function and the outlook of individuals with MS, as well as their relationship with their spouse and family (Horton et al., 2015).

The six studies discussed regarding resources have provided evidence on the positive impact this factor has regarding adjusting and adapting to a diagnosis of multiple sclerosis in adults with RRMS. Physical, financial and practical support and assistance from family, friends, and healthcare professionals as well as the personal involvement in self-help groups and interventions were cited to increase relationships among family members, improve family dynamics, and promote family resilience and a since of return to families having a normal life.

Literature Review on Family Appraisal

When families are faced with a loved one who is diagnosed with MS, family members appraise the seriousness of the diagnosis for the family. Families can appraise the meaning of the diagnosis as well as the family situation that the diagnosis imposes. This includes assessing demand and responsibilities. In addition, families can appraise the diagnosis based on their family schema, which are their family's shared values, beliefs, rules and goals. Five empirical studies were included in the review of literature regarding family appraisal and RRMS and are listed in the table in Appendix E. Four of these five studies, which were all qualitative, were incorporated in supporting literature regarding factors of demands and/or stressors as well

(Bjorgvinsdottir & Halldorsdottir, 2014; Bogosian et al., 2009; Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014). The one study that has not been previously mentioned in this review is a mixed methods study that involved next of kin of individuals diagnosed with MS (partner, spouse, parent, sibling, or adult child) (Liedstrom, Isaksson, & Ahlstrom, 2010) .

All five of the studies reviewed agreed that the situational appraisal of the diagnosis initially had negative connotations. In a study of 11 young caregivers of parents diagnosed with MS, the caregivers, who ranged from age 5 to 18, viewed their caregiving role and the direct consequences of the care as "silent, invisible and unacknowledged" (Bjorgvinsdottir & Halldorsdottir, 2014). In one of the four other studies, the caregivers viewed providing care to a person with MS on as having a negative impact on their own social life (Bogosian et al., 2009). In another study of 44 next of kin of individuals diagnosed with MS, the family appraised health related quality of life as a direct indicator of being healthy and having the freedom to fulfill their life desires (Liedstrom et al., 2010). The next of kin viewed MS symptoms as a sign of decreased quality of life that caused a decreased in freedom, self-actualization and security, leading to negative a negative emotional experience (Liedstrom et al., 2010).

Two of the five studies discuss family appraisal based their shared beliefs, values, rules and goals (schema) and how it shaped their perceptions about MS. In one of the qualitative studies examining the experiences of individuals who are confronted with their diagnosis of MS, the initial appraisal of diagnosis is based on the false perceptions and inadequate information that they have about the disease, its course and clinical manifestations (Fallahi-Khoshknab et al., 2014). Bostrom & Nilsagard (2016) discuss the

"prerequisites within the family" in which both the individual history and family history of their understanding and thoughts about MS shape their outlook and reaction to the diagnosis.

The five studies reviewed regarding family appraisal provided evidence on the impact this factor has regarding adjusting and adapting to a diagnosis of multiple sclerosis in adults with RRMS as well. The initial appraisal of false perceptions among families of adults diagnosed was due to their personal and family's lack of understanding about the disease process and its clinical manifestations. The supporting evidence in this review of the literature that their outlook and reactions to the diagnosis shaped by the family's initial appraisal. Furthermore, this review indicated that situational appraisal of families and appraisal based on the family's schema has a direct impact on what they perceived as quality of life, freedom, and self-actualization.

Literature Review on Family Problem-Solving Communication and Coping

The literature reviewed regarding family problem-solving communication and coping are two concepts intertwined in the constructs of the Resiliency Model. When families are confronted with the demands of a chronic illness such as MS, they must learn to organize these stressors into a manageable course of action that will constructively restore harmony and balance (McCubbin, Thompson, & McCubbin, 2001). Problem solving communication and coping are two methods by which balance and harmony can be achieved. Affirming communication is the most effective way for families to adapt to situations that may cause stress and strain (McCubbin, Thompson, & McCubbin, 2001). The literature supported affirming communication as the most optimal method of communication when families began to problem solve in the presence of stress and strain

(Bostrom & Nilsagard, 2016; Ghafari, Fallahi-Khoshknab, Nourozi, & Mohammadi, 2015; Paliokosta et al., 2009). With affirming communication, active listening is involved without physical or emotional harm to the persons involved in conversation to end conflict on a positive note. Because of communication and well-organized problem solving, effective coping began to take place (Bogosian et al., 2009; Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014).

Eleven empirical studies were included in the review of literature regarding problem-solving communication and coping and RRMS and are listed in the table in Appendix F. The review included eight qualitative studies and two quantitative studies and one mixed-methods study. Six of the eight qualitative studies mentioned in a previous section of this literature review (Bogosian et al., 2009; Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014; Ghafari et al., 2014; Malcomson et al., 2008). The two remaining qualitative studies included individuals with MS and their spouses (Boland, Levack, Hudson, & Bell, 2012) and adolescents with a parent with MS (Mauseth & Hjälmhult, 2016). The two quantitative studies not yet discussed involved the participation of families, as a unit, with the individual with MS, a spouse or partner and one child or offspring of the person with MS (Ehrensperger et al., 2008; Paliokosta et al., 2009). The mixed methods study not previously mentioned involved individuals with MS only (Mikula et al., 2014).

Seven of the eleven studies examined coping exclusively (Boland et al., 2012; Ehrensperger et al., 2008; Fallahi-Khoshknab et al., 2014; Malcomson et al., 2008; Mauseth & Hjalmhult, 2016; Mikula et al., 2014; Turpin et al., 2008), while one study examined coping and communication (Bostrom & Nilsagard, 2016) and another included

coping and problem solving (Ghafari et al., 2014). One study included communication exclusively (Paliokosta et al., 2009) and problem solving exclusively (Bogosian et al., 2009).

The literature review on coping and problem-solving communication after a MS diagnosis will be discussed based on the following four categories of participants: 1) the individual with MS exclusively; 2) the dyad couple; 3) adolescents; and 4) the family as a unit. Participants in three of the 11 studies involved individuals exclusively (Fallahi-Khoshknab et al., 2014; Malcomson et al., 2008; Mikula et al., 2014). Five of the 11 studies involved the dyad couple as participants (Bogosian et al., 2009; Boland, Levack, Hudson, &. Bell, 2012; Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014; Ghafari et al., 2014). Two of the eleven articles reviewed involved adolescent children of individuals with MS as the participants (Mauseth & Hjalmhult, 2016; Turpin et al., 2008). One of the eleven studies involved families as a unit and included persons with MS, their spouses, and a child within the family (Ehrensperger et al., 2008).

Three of the seven studies regarding coping that involved individuals with MS reported that coping is more closely associated with emotional support than physical support (Fallahi-Khoshknab et al., 2014; Malcomson et al., 2008; Mikula et al., 2015). After individuals with MS begin to accept the diagnosis, they can begin to problem solve and cope effectively. According to Mikula, et al. (2014), coping began by stopping unpleasant emotions and thoughts about the disease (Mikula et al., 2014). According to Malcomson (2008), coping meant “getting on with day to day life” (Malcomson et al., 2008). Coping for individuals with MS involved being proactive about their health and well-being, enhancing their knowledge about MS, learning their realistic limitations,

changing their belief systems and learning how to juggle tasks that they need to perform (Malcomson et al., 2008). The third study involving 25 individuals with MS, although limited by a small sample size, alluded to the fact that individuals with MS turn to their faith in God and submit to him for peace (Fallahi-Khoshknab et al., 2014). Malcomson et al (2008) concluded that successful coping is developed from personal resources such as peer support rather than any other support, although patients rely on health professionals to provide guidance regarding personal needs and access to experts that provide insight into coping and self-management.

The five studies regarding coping and problem-solving communication that involved dyad couples revealed that coping among couples developed over time. Boland et al (2012) refers to couples coping with MS as “peaks and troughs”. Two of the studies reported that coping occurs daily (Bogosian et al., 2009; Boland et al., 2012). These two studies concluded that coping among couples includes renegotiated and shared responsibilities, adjusted and reorganized activities, normalization and acceptance of challenges that arise and implementation of solutions to practical problems (Bogosian et al., 2009; Boland et al., 2012). Two additional studies added that when an individual is diagnosed with MS that individual and his/her partner perceive themselves as a unit, with a sense of purpose and togetherness who rely on emotional support and an open and comfortable relationship (Boland et al., 2012; Ghafari et al., 2014). Boland et al. (2012) discussed in his findings that if coping styles were different within the couple, then there was an increased likelihood of conflict, frustration and opposition (Boland et al., 2012). Within this relationship, the spouse of the person with MS enabled communication about problems, expectations and needs. Boland et al. (2012, p. 1370) added that coping

"occurs over the long haul" and incorporates the concept of hope and faith in self and faith in each other.

The two studies regarding coping and problem-solving communication that include adolescent participants gave insight into the differences in coping needs based on developmental capabilities (Bostrom & Nilsagard, 2016; Turpin et al., 2008). In one of the studies on adolescents who functioned as caregivers of the parent, effective adolescent coping was achieved when: 1) adolescents could express their emotions; 2) adolescents sought social support; 3) adolescent were allowed time for recreation and 4) were involved in the diagnosis (Turpin et al., 2008). The findings from Bostrom et al (2016) agreed that effective coping involved open communication with the family in which the children and other family members are involved. This study also revealed that a lack of communication within the family or between health care professional and the family can put a strain on one or more of the family members (Bostrom & Nilsagard, 2016).

The one study that involved the participation of families as a unit concluded that the individual with MS' ability to cope is associated with their emotional and neuropsychological function and not their physical disability (Ehrensperger et al., 2008). This finding was supported in three studies focused solely on the person with MS. (Fallahi-Khoshknab et al., 2014; Malcomson et al., 2008; Mikula et al., 2014).

Eleven studies were discussed regarding factors of communication, coping, problem solving all of which were intertwined in the literature regarding individuals and their families with MS. The discussion of these factors was approached by a discussion of the individual, couples, adolescents and the family as a unit. Individual coping was

associated more with emotional support that was emotionally personal rather than physical which increased the person with MS level of acceptance and their ability to problem solve and cope. The literature on couples provided evidence that coping and problem-solving communication occurred over time with the couples functioning as a unit and that togetherness of the couples provided emotional support and decreased conflict and opposition. Regarding the literature on adolescents, effective coping was evident when adolescents could express their emotions, provided social support, allowed time for self and involved in the diagnosis of a loved one with MS. The literature on the family as a unit concluded that open communication within the family was necessary to prevent strain on the members.

Literature Review on Family Adaptation

Adaptation is characterized by the balance and harmony of the family's relationships, the family's function and the family's developmental well-being as a unit of individuals who are connected emotionally and spiritually within their own natural environment as well as within the community (McCubbin, Thompson, & McCubbin, 2001). It is the stability and balance of these connections that bring challenge and ultimately result in solutions to those challenges (McCubbin, Thompson, & McCubbin, 2001). The pile up of demands are on one end of the spectrum and societal and family resources, positive appraisal, effective problem-solving communication and coping on the other (McCubbin, Thompson, & McCubbin, 2001). The review of literature revealed only four studies that address the adaptation of people living with RRMS and is listed in table in Appendix G (Bostrom & Nilsagard, 2016; Ghafari et al., 2015; Hwang, Cvitanovich, Doroski, & Vajarakitipongse, 2011; Starks, Morris, Yorkston, Gray, &

Johnson, 2010). Two of the four studies that discussed the various aspects of adaptation were mentioned previously in other portions of the literature review as these studies also pertained to other concepts (Bostrom & Nilsagard, 2016; Ghafari et al., 2014). Of the two studies that have not been previously mentioned, one is a quantitative study (Hwang, Cvitanovich, Doroski, & Vajarakitipongse, 2011) and the other one is mixed method study (Starks, Morris, Yorkston, Gray, & Johnson, 2010). Two of the four studies identified the adaptation of individuals to a diagnosis of MS (Ghafari et al., 2015; Hwang et al., 2011). One of the four studies identified the adaptation of couples to a diagnosis of MS in one partner (Starks et al., 2010). The last of the four studies identified the adaptation of family as a unit (Bostrom & Nilsagard, 2016).

Two of the studies on the adaptation of individuals with MS discussed strategies that lead to the adaptation to MS (Ghafari et al., 2015; Hwang et al., 2011). One of these studies identified three factors that enriched the adaptation of individuals with MS and correlated conversely with positive quality of life (Hwang et al., 2011). These factors included self-concept, social support and accessibility (Hwang et al., 2011). Positive self-concept was directly related to the person regaining a sense of personal well-being (Hwang et al., 2011). The study also concluded that physical, psychosocial and spiritual adjustments to values and beliefs as well as relationships to others and to one's surroundings must change over time in order for adaptation to take place (Hwang et al., 2011). Accessibility to quality and reliable health care in the presence of disability correlated with positive quality of life and effective adaptation (Hwang et al., 2011).

The other study on the adaptation of individuals with MS revealed seven themes which included: religiosity, information seeking, emotional reactions, concealing the

disease, maintaining hope, seeking support, and fighting the disease (Ghafari et al., 2015). Religiosity which refers to the various aspects of religious activity, belief or commitment, was demonstrated by the participant's trust in God and in prayer as a means to achieve peace and acceptance (Ghafari et al., 2015). Adaptation through information seeking was achieved by individuals' research and education (i.e. attending seminars, internet, brochures, etc.) about a disease that they once knew nothing or little about in order to improve their knowledge (Ghafari et al., 2015). Some individuals with MS resorted to emotional reactions of anger, sadness and crying so that they could relieve themselves of those emotions and move on (Ghafari et al., 2015). Yet, other individuals with MS concealed the disease because they perceived that others would pit them or reject them as a person (Ghafari et al., 2015). The individuals with MS also had a fear of unemployment (Ghafari et al., 2015) . The study revealed that because of the individual with MS and their social interaction and efforts toward reaching personal goals, they began to adapt and maintain the element of hope (Ghafari et al., 2015).

In addition to the seven themes identified, the study revealed that unmarried participants received emotional support of sympathy, comfort and protection from parents and friends while married participants received support of empathy and active listening from their spouses (Ghafari et al., 2015). Physical support from significant others (family, spouses and friends) included transportation to therapy sessions and medical care and assistance with activities of daily living (Ghafari et al., 2015). Financial support from state agencies such as the MS society were reported by participants who had lost their jobs due to the illness and who experienced high cost of medical treatments (Ghafari et al., 2015). However, participants reported a need of support from the

government to update public facilities that are not handicap accessible and areas such as sidewalks that are not safe as well as the government's active assistance in finding suitable jobs for individuals with disabilities (Ghafari et al., 2015). Lastly, fighting the disability and the disease was an important strategy in which individuals with MS made changes to their behaviors and lifestyles, used medications and complementary and alternative therapies, and maintained their functional abilities (Ghafari et al., 2015).

In couples' research regarding adaptation in MS, Stark et al (2010) identified two patterns of couple adaptation: "in sync" and "out of sync". According to the study, couples are "in sync" when the partner diagnosed with MS and the partner without MS have compatible world views and communication strategies and are able to respond collectively to solve problems and challenges (Starks et al., 2010). On the other hand, couple who adapt and are "out of sync" with each other, usually adjusted to change at a different pace and used strategies with different priorities and goals regarding parenting and medication selection for treating MS as their focus, which in turn caused additional strain and maladaptive behavior within the relationship (Starks et al., 2010). The "out of sync" couples had differing personality styles and the shifts in their newly appointed roles caused even greater tension. Of note, this was the only study that utilized a theoretical model, employing Patterson' s Family Adjustment and Adaptation Response (FAAR) model as a guide in the development of the identified themes to describe the adjustments and adaptations for couples. The Resiliency Model of Family Adjustment and Adaption, which is the underpinning of the proposed study, was derived from previous adjustments to the FAAR for better understanding of the role of coping in adjustment and adaptation (McCubbin & Patterson, 1983).

The last study in the review of literature regarding adaptation described the family as a unit, in which all members of the family have their own history and developmental phase and react in their own way (Bostrom & Nilsagard, 2016). Bostrom et al (2016) summed up the idea of adaptation when it came to a person with MS by stating that MS is a "family matter." The use of separate focus groups with ill parents, healthy parents and children of a diagnosed patient concluded that all family members needed to be recognized in the face of chronic illness while adapting to everyday life and developing strategies to manage the disease as a family (Bostrom & Nilsagard, 2016).

Four studies were discussed regarding family adaptation of a person who has been diagnosed with MS and their family. The literature cited in this portion of the review identified the needs of individuals, couples and families to adapt to the diagnosis of MS to improve the quality of life of all parties involved. Individual needs included the identity of self, social support and accessibility. In addition, the individual person and his significant others need to change their values and beliefs to move toward adaptation of MS because of the progressive nature of the disease. Other factors that were identified that influenced adaptation included, religion, information seeking, maintaining hope, seeking support, access to quality and reliable healthcare, being in-sync with your partner, and fighting the disease. Finally, recognition of the family and the integral part that it plays as a unit in the process of adaptation is a big part in the process and the success of any of the strategies that families put into place.

Summary of the Integrative Review

The purpose of this integrative review was to examine the experiences of individuals and their families in the adaption to RRMS and the impact that factors such as

demands, resources, appraisal, problem solving communication and coping may have on this process. The findings of 26 empirical studies were reviewed. Each of the factors was reviewed individually to determine which the factors supported the construct of adaptation. Some of the studies revealed knowledge on multiple concepts and this was discussed as well. Based on the theoretical underpinning of the proposed study, the studies reviewed support that the balance of negative factors such as stress and strain and positive factors such as coping, social support, problem solving, resources will result in the move toward adaptation over time.

Thirteen empirical studies regarding the concept of the demands placed on individuals and their families were discussed in the review. The literature provided knowledge of the demands on individuals and their families who were recently diagnosed with RRMS (Aymerich et al., 2009; Bogosian et al., 2009; Fallahi-Khoshknab et al., 2014; Heward et al., 2006; Koopman et al., 2006; Malcomson et al., 2008). The knowledge included: initial perceptions of the diagnosis, sense of helplessness, loss of control, knowledge deficit about the disease and its clinical manifestations and outcomes, uncertainty of the future, fear of job termination, decreased quality of life, and social isolation, as well as lack of support and understanding from family, friends and social networks (Aymerich et al., 2009; Bogosian et al., 2009; Fallahi-Khoshknab et al., 2014; Heward et al., 2006; Koopman et al., 2006; Malcomson et al., 2008). In addition, the needs of the affected families because of these demands were also identified and included professional support, interpersonal relationships, adjustments to current employment, personal support, maintenance of social life and control over personal life. Resolving these needs would enable diagnosed individuals to cope with the new circumstances

within the family and society (Heward et al., 2006; Koopman et al., 2006; Malcomson et al., 2008). The literature also provided specific knowledge of how the role of parenting, relationship and responsibilities of children are affected by a parent who is diagnosed with MS were addressed (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016; Diareme et al., 2006; Pakenham & Cox, 2012; Steck et al., 2007; Turpin et al., 2008).

Six empirical studies were reviewed regarding the concept of resources as it relates to individuals with RRMS. This portion of the review revealed a list of resources such as physical care and support, financial support and practical assistance that were utilized to balance the effects of the demands on individuals with MS and their families (Diareme et al., 2006; Ghafari et al., 2014; Pakenham et al., 2012). The review of literature on resources also revealed additional resources that function as internal motivation such as self-help groups, and self-management programs that changed the outlook of individuals with MS and their families (Eliášová et al., 2015; Horton et al., 2015; Mulligan et al., 2016).

Additionally, five empirical studies were reviewed regarding the concept of appraisal. Findings revealed that situational appraisal began initially with negative connotation for caregivers and next of kin (Bjorgvinsdottir & Halldorsdottir, 2014; Liedstrom et al., 2010). Situational appraisal was supported by the personal insights and false perceptions and misinformation of those individuals who were diagnosed with the disease (Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014).

Because the concepts of family problem solving communication and coping are interconnected, the literature regarding these two concepts was reviewed collectively.

Eleven empirical studies were reviewed regarding these two concepts. This portion of review revealed problem solving communication and coping to include the family, within a dyad relationship, and as an individual with MS. Coping was the result of emotions in which individuals began the process of accepting their diagnosis and moving on with their lives with the personal support of their peers, family, caregivers (Bogosian et al., 2009; Boland et al., 2012; Ehrensperger et al., 2008; Fallahi-Khoshknab et al., 2014; Malcomson et al., 2008; Mikula et al., 2014). Effective coping occurred in the presence of problem solving communication during which the individual with MS had an opportunity to express their problems, needs and expectations of others as well as the recognition of the effects that MS has on the family (Boland et al., 2012; Ghafari et al., 2014; Turpin et al., 2008).

Lastly, four empirical studies on adaptation in individuals with MS and their families were reviewed. Strategies that influence the adaptation of individuals with MS included developing self-concept, having accessibility to care, seeking support, maintaining hope, religiosity, information seeking and fighting the disability and disease (Ghafari et al., 2014; Hwang et al., 2011). Patterns of couple adaptation included working together as a team and having the sharing the same goals as a couple to achieve the best outcome and adaptation within the family included the recognition of the needs of each of the family members while developing strategies of adaptation to MS (Bostrom & Nilsagard, 2016; Starks et al., 2010).

Strengths and Limitations

To the author's knowledge, this is the first integrative review of literature to include empirical research from quantitative and qualitative studies examining factors

that influence adaptation of RRMS in adults. This review confirms that adaptation is influenced by many factors and is based on an individual's firsthand experiences. The review uncovered that demands can be based on the individual with MS, within a dyadic couple, on the child or loved one of an individual with MS or on the family as a unit. Resources can be individual and collective and complement an individual's appraisal of the diagnosis, the family's appraisal of the situation a as result of the diagnosis and the collective problem-solving communication and coping skills to achieve balance.

` This review uncovered limitations and strengths of the published articles, as well as gaps in the literature. More than half of the articles (N=15) that were reviewed regarding factors that influence adaptation were from the perspective of the individual which included those with MS, partners of individuals with MS or children of individuals with MS. Nearly one-third (N=6) of the articles reviewed were from the perspective of a dyad between the individual with MS and a spouse or another member of the family, but only 5 of the 26 articles were from the perspective of the family as a unit. Because of the clinical manifestations of the disease, MS is a disease that needs to be examined from the perspective of the family as a unit. Factors that affect the individual may influence how the family copes and adapts to a diagnosis and future empirical research in this area will support the need for family interventions on coping strategies. Half of the 26 studies (N= 14) involved qualitative studies with methodology that would produce rich and in-depth information. This information from these studies was collected by means of semi-structured or unstructured interviews, as well as open-ended questions that were collected individually or through focus groups. Of those 14 studies reviewed, only one involved the collective information of the family as a unit.

One third of the studies involved in this review of literature were from quantitative studies that utilized large data sets, which makes the information gathered more generalizable. Three of the studies involving data sets of 50 to 100 individuals with self-reported validated measures as a means of gathering supporting data for factors that may influence adaptation such as quality of life and emotional and behavioral problems (Diareme et al., 2006; Eliášová et al., 2015; Hwang et al., 2011). Three of the nine quantitative studies in this review involved large samples of dyads involving the individual with MS and their caregiver, spouse or partners using self-reported questionnaires (Aymerich et al., 2009; Koopman et al., 2006; Labiano-Fontcuberta et al., 2015). Four of the nine quantitative studies reviewed involved the family as a unit and involved the use of self-reported, validated questionnaires on the psychological consequences of the disease on the child and parent, the coping skills of the family and issues of communication (Diareme et al., 2006; Ehrensperger et al., 2008; Paliokosta et al., 2009; Steck et al., 2007). Although this method of data collection can be powerful and can yield information that can be readily analyzed, this method of data collection may result in response bias, which may skew the true findings of the study (Polit & Beck, 2012). In addition, the review of literature provides a summary of the factors that may influence the adaptation of the individuals and families living with MS. However, only one article that presented all the factors collectively so that the reader could better understand how adaptation occurred over time.

An important gap in the literature is the lack of empirical research regarding diagnosed individuals less than 18 years of age and the adaptive process that occurs with these individuals and their families, although there is literature to support that children

are being diagnosed with MS with variable clinical manifestations (MacAllister et al., 2009). There may be differences in how families adapt to a child with MS in comparison to how families adapt to an adult diagnosed with MS. The demands and challenges within the relationships of those families of children with MS has yet to be explored. Because of the rarity of pediatric onset MS, the resources may be different for families of children with MS than those resources identified in adults with MS and their families. Because of the uncertainty of the future, families of children with MS may appraise the diagnosis differently from those families of adults with MS. Problem solving communication and coping in the face of a life-long illness without a cure may prove to be difficult. The unpredictability of the disease along with the increased disease activity in the initial stages of the disease in pediatric MS may be the source of delayed adaptation or maladaptation, but this is unknown as well.

Examining Family Adaptation in Juvenile Idiopathic Arthritis

Due to the lack of research on family adaptation to POMs, this section presents three seminal articles regarding adaptation of families to a similar chronic illness with pediatric onset juvenile idiopathic arthritis (JIA). These studies will be used to guide the methodology of the proposed study to compare the adaptation of families to a diagnosis of JIA to the adaptation of families to a diagnosis of POMS. Prior to the review of the seminal studies, a brief summary of the etiology of JIA will be presented.

Brief Overview of JIA

JIA is like POMS in numerous aspects. JIA, the most common rheumatological disease in children, is a broad diagnosis that covers all forms of arthritis in children

before the age of 16 (Ravelli & Martini, 2007). Just as with POMS, JIA may cause both short term and long-term disability, its onset is unpredictable, and it is without a cure (Ravelli & Martini, 2007). The prevalence of both conditions is similar and the etiology of both conditions is unknown (Lawrence et al., 1998). Children with any form of JIA can experience acute and chronic episodes of pain, inflamed joints, decreased mobility, contractures, and visual problems as well as growth delay (Ravelli & Martini, 2007). Like POMS, the treatment options are usually medications that cause undesirable side effects. As with POMS, JIA has the potential to disrupt family functioning due to the feelings of uncertainty and loss of control in addition to the added responsibilities, daily challenges and demands of family members of a child with JIA. (Moorthy, Peterson, Hassett, & Lehman, 2010).

Review of Seminal Articles on the Adaptation of JIA

One seminal study on family adaptation to JIA was conducted in the early 1990s and the other two were conducted in the late 1990s. In the first study, a stress and coping model were tested using a repeated measure design and longitudinal analysis with 159 married couples at Time 1 and 111 of these couples one year later at time 2 (Timko, Stovel, & Moos, 1992). The purpose of the study was to examine the adaptation of mothers and fathers of children with juvenile rheumatological disease faced with ongoing life stressors, child and parental dysfunction while utilizing family resources and parental illness- related coping skills (Timko et al., 1992). Analysis revealed that psychosocial problems in children were linked to maternal depression, strain caused by the disease, and lower sense of managing stress.

Frank et al (1998) compared 107 children with JIA to 114 children with Type 1 Diabetes and 88 healthy controls on a battery of self-reported and validated measures of child behavior and emotional function, as well as on coping and other psychological functions of their parents. (Frank et al., 1998). This study used the Risk and Resistance Model, a broad model of adaptation in which the effect of any risk factor is variable and dependent on other buffering risk factors such as family environment (Frank et al., 1998). This study concluded that children with chronic disease experience emotional and behavioral problems related to parental distress and not related to their specific medical diagnosis (Frank et al., 1998). These conclusions support the hypothesis that children with chronic illness in the presence of risk factors such as maternal depression and greater disease activity will demonstrate more adaptive difficulties and promote family dysfunction (Frank et al., 1998).

The last of the three studies was conducted in 1999 with 30 adolescents diagnosed with JIA and their family members (Degotardi, Revenson, & Ilowite, 1999). This study on the adaptation of families of children with JIA had a two-fold purpose. One purpose was to describe how family function is disrupted or changed by JIA and to identify the coping strategies used to deal with JIA related stressors by adolescents and their families (Degotardi et al., 1999). The study's second purpose was to explore the use of interviews to describe the findings of the study. In addition to a structured interview, participants completed two self-report validated measures and the Youth Self Report and Family Environment Scale (Degotardi et al., 1999). The results of the study revealed that families of children with JIA used three types of coping strategies: appraisal, problem-focused and emotion focused (Degotardi et al., 1999). Problem focused coping was

related to treatment adherence and how the families dealt with this issue. For example, those families that came up with very specific routines and mutually agreed upon methods to encourage adherence improved their level of coping strategies within the family. Emotion focused coping such as impulsive outbursts, on the other hand, were indicators of poor adjustment, evidence of greater family conflict and lack of family cohesion (Degotardi et al., 1999). Optimistic appraisal were correlates of improved adjustments in families affected by JIA and pessimistic appraisals were indicators of poor functioning (Degotardi et al., 1999). Study conclusions identified that qualitative and quantitative data gathered using interviews can be useful in assessing family level coping and adaptation.

The three seminal studies reviewed on JIA will help to guide the interpretation of the finding of the proposed study on POMS. Timko and colleagues (1992) provided insight regarding the importance of gathering data over time to validate ongoing stressors a family may face when dealing with chronic illness and how these issues may be linked to stressors that may cause families to maladapt. Frank and colleagues (1998) provided information on the influence of parental distress on the emotional and behavioral problems experienced by the child with chronic illness unrelated to the diagnosis (Frank et al., 1998). Lastly, Degotardi and colleagues (1999) provided insight on family appraisal, problem focused coping and emotion focused coping, all of which could influence the outcome of adaptation in families of children with chronic illness. (Degotardi et al., 1999). The review of these three studies on pediatric patients will a chronic illness, as POMS along with the literature from adults with RRMS will drive the discussion and analysis of the proposed study.

Summary

Chapter 2 provided a detailed overview of MS, which included the history, etiology, disease course, clinical manifestations, diagnostic criteria and current treatment options. This overview provided a comprehensive examination of the epidemiology of MS to facilitate understanding of the factors that may be involved with adjustment and adaptation to a diagnosis. Next, the theoretical framework, the Resiliency Model of Family Stress, Adjustment and Adaptation, including its concepts and relevance to the subject were introduced to the reader. This provided an outline so that the review of literature would have direction and it presented details of the theoretical framework as it will use to guide the development and analysis of the proposed study. An integrative review of the literature identified concepts and the major constructs of the theory under investigation, adjustment and adaptation. This review covered factors such as demands, resources, appraisal, problem-solving communication and coping of adults with RRMS.

The literature review included articles on adaptation to RRMS but does not link all the factors that may influence the process of adaptation. Furthermore, the literature lacked empirical documentation of the factors that affect familial adaptation. Three seminal studies regarding the adaptation of families to juvenile idiopathic arthritis (JIA) were included as this condition is similar in etiology, clinical manifestations, prognosis, and treatment options and has been researched regarding family adaptation. It was evident by the literature on JIA that other family factors such as parental distress, maternal depression, stress caused by the disease and lower sense of managing stress as well as coping strategies (optimistic versus pessimistic) problem focused coping and emotion focused coping can influence adjustment and adaptation which faced with

chronic illness in families of children with POMS. These studies will be utilized along with the Resiliency Model of Family Stress, Adjustment and Adaptation to shape the analysis of the proposed study along with the regarding the factors that influence the adaptation of families of children with POMS, due to a significant gap in the literature. A qualitative descriptive study will capture the essence of this phenomenon, providing insight into ways in which families can maintain a balance between demands and harmony. This knowledge is needed to develop interventions aimed at families of children with POMS that will offer coping strategies to increase positive adaptation over time. Findings in this study will contribute knowledge to the literature that has never been explored before on this topic. The next chapter will provide a detailed and in-depth description of the research design and methodology that will answer the research questions for the proposed study.

CHAPTER 3

METHODS

In this chapter, a discussion of qualitative research inquiry and the methodology and rationale for the selection for this study are included. The design of the study is then described in-depth to include the sampling strategy, recruitment plan, study setting as well as data collection procedures and analysis. The chapter will conclude with strategies for ensuring reliability and validity of the study as well as the protection of human subjects.

Specific Aims and Research Questions

The research study was guided by the following central research question:

> How do family factors influence adaptation in families of children diagnosed with multiple sclerosis?

The central question was further supported by the following aims and research sub-questions:

Aim 1: Explore the initial and ongoing psychosocial, financial, emotional and behavioral impact on families who have a child with POMS.

- What are the stressors and strains experienced by families of children with POMS?
- What are the strengths and capabilities of individual family members, the family working as a unit and within the community?

Aim 2: Explore how the family adjusts to raising a child with POMS

- How do caregivers of children with POMS appraise or view their experience of raising a child with POMS?
- What are caregiver's perceptions of their ability to manage the demands associated with the medical management of a child with POMS?

Aim 3: Explore caregivers' perceptions of family's strategies for coping and balancing the needs of the child with POMS and of the family.

- What strategies does the family use to cope with the ongoing challenges associated with raising a child with POMS?
- Regarding immediate family members, how well are families of children with POMS able to achieve a balance between the needs of the child with POMS, the needs of the family as a whole and the needs of other family members within the home?

Aim 4: Identify resources that families perceive are available to assist families caring for a child with POMS.

- How do caregivers describe the availability of resources to assist families' adaptation to raising a child with POMS?

Methods of Qualitative Inquiry

There are five traditional approaches to qualitative inquiry: narrative, phenomenology, grounded theory, ethnography and case study. Narrative research is based on the experiences of individuals and their told stories and how they perceive

themselves (Hughes, Locock, & Ziebland, 2013). Phenomenology research describes the common meaning (essence) of individuals (up to ten participants) as they live their experiences within a phenomenon (Creswell, 2013; Strickland, Worth, & Kennedy, 2015). Grounded theory moves a step further than describing the essence of the phenomenon into discovery and generation of theory to explain the actions and processes of the participants (Foley & Timonen, 2015). Ethnographical research, on the other hand, examines shared patterns of values, beliefs, behaviors and language that may be common to study participants that are not likely located in the same place but share a common culture (Polit & Beck, 2012). Ethnography is characterized by long-term observation where the researcher spends extended time in a particular social group in order to collect data (Morgan-Trimmer & Wood, 2016). Finally, case study is an in-depth investigation of a single entity or small number of entities (individual, family, group, institution, community, or other social unit) in which a detailed description is gathered and the relationships among the phenomenon are examined over time (Polit & Beck, 2012; Sandelowski, 2011).

Qualitative descriptive studies are rarely discussed among the traditional approaches although there is a strong suggestion that many nursing studies that involve qualitative data collection and analysis are purely qualitative descriptive studies that are not linked to any particular tradition. Polit & Beck (2009) conducted an analysis of more than 1,000 studies that were published in eight journals between 2005 and 2006. The analysis found that more than half of these studies (52%) were qualitative descriptive studies that utilized broad methods of content analysis rather than a formal system of analysis associated with one of the five traditional approaches. Sandelowski (2000)

noted that this form of data collection and analysis presents an interpretative and descriptive summary of a phenomenon. Nearly a decade later, Sandelowski revisited the concept of qualitative description to clarify that the method should be used as a means for presenting research that resists simple classification but not to name poorly conceived and conducted studies after-the fact (Sandelowski, 2010).

Study Design

The research design for this study was a qualitative descriptive inquiry. The major underpinning of qualitative research, which is supported by constructivist tradition, is the complex nature of humans and their innate ability to create and shape their own experiences and realities (Polit & Beck, 2012). Qualitative research is heavily focused on understanding these experiences as they are lived through in-depth probing and interpretation of narrative and subjective material provided by its participants. These experiences may be hard to quantify with quantitative methodologies (Sandelowski, 2000). Oftentimes, qualitative researchers study an unknown phenomenon or a phenomenon about which little is known to seek clarity or further identification regarding its existence (Sandelowski, 2000). Qualitative approaches are also an excellent way to investigate family dynamics and family relationships because the researcher can explore meanings of an phenomenon unique to families (Ganong & Coleman, 2014) .

Qualitative descriptive research allowed the researcher's exploration of factors that influenced families' adjustments and adaptation to the diagnosis of POMS. This unexplored phenomenon affected the child diagnosed with POMS as well as the entire family. In line with the purpose of the study, in-depth interviews were conducted to

identify universal commonalities among families during the analysis phase to provide insight into the development of family interventions.

Site of Participant Recruitment

The primary site for recruitment of caregivers of children diagnosed with POMS was the Center for Pediatric Onset Demyelinating Disease (CPODD) located at Children's Hospital of Alabama (CHA). CPODD is one of the six original nationally recognized Pediatric MS Centers of Excellence established by the National Multiple Sclerosis (MS) Society in 2006 in an effort to address the needs of children and families who are diagnosed with rare disorders of the central nervous system such as MS (National MS Society, 2007). Since its inception, the center has seen over 135 children and adolescents diagnosed with POMS and another 100 children who have presented with a single demyelinating event who may go on to develop POMS in the future. On average, six patients previously diagnosed with POMS and between one and two patients newly diagnosed with POMS are seen monthly at the center. POMS patients seen at the center are disproportionately female (68%), nearly half are African American (41%), and the mean age of MS onset is 13.1 (SD = 4) years. Most of the patients and their families fall in the median socio-economic status. Children and adolescents with POMS come to CPODD from the surrounding metropolitan area as well as from the entire state of Alabama, and surrounding states including Georgia, Mississippi, Tennessee, Florida, Louisiana, North Carolina, South Carolina and Arkansas. The center was the optimal place to gain access to potential participants for this study.

Sampling Strategy

Purposeful sampling was used for selecting potential research participants for the study. Purposeful sampling is used in qualitative research for the selection of participants who will provide rich cases when resources are limited (Palinkas et al., 2015). More specifically, criterion sampling, a specific form of purposive sampling that involves the selection of participants based on a pre-determined criterion of importance, was used as it works well to provide a representation of the population and their phenomenon to be studied (Creswell, 2013). The participants had to meet the following inclusion criteria for this study: 1) be a primary family caregiver of a child who has been diagnosed with MS as defined by the 2010 McDonald Criteria and the 2013 International Pediatric MS Study Group 2) be 18 years of age or older, 3) have a child diagnosed with a POMS diagnosis (for at least 6 months who is $\leq$ 22 years of age) and who has received previous or on-going care and/or consultation in the last year at the CPODD, 4) be able to read and speak English, 5) provide 50% or more of the care to the child with POMS, 5) live in the household with the child with POMS. Participants were excluded for the following reasons: 1) the primary language is other than English and 2) the child has not been diagnosed with POMS according to the diagnostic criteria but is being treating with a disease modifying therapy. Caregivers were targeted for this study, as they would be able to provide the most information about the patient and family structure and dynamics. In addition, the researcher decided to seek out caregivers of children who had been diagnosed for at least six months with the hope that participants and their families would be over the initial shock of a diagnosis and treatment would have been initiated.

Once the UAB Institutional Review Board (IRB) granted study approval, the electronic medical records were queried for patients with a diagnosis of multiple sclerosis and then the potential participants were screened using a questionnaire to establish qualifications based on the inclusion/exclusion criteria (see Appendix H for screening tool). The names, phone numbers and mailing addresses of all potential participants were obtained from the hospital medical record and were recorded for recruitment purposes. The potential participants meeting the criteria for participation were mailed an informative flyer regarding the study as well as a letter from the treating physician at the center supporting the study to be conducted (see Appendix I for flyer and Appendix J for letter of support). This method of passive recruitment included local as well as out of town participants to ensure the richness of information of participants. The flyer included the following information: study purpose, study location, researcher contact, study benefits, confidentiality, disclosure of results, study convenience, the right to withdraw without penalty and the potential for a follow-up interview.

Families with upcoming appointments, who were not screened in the initial screening process, were screened prior to their appointment to identify other potential participants. If any of the families meet the screening criteria, active recruitment over the phone occurred three days prior to a potential participant and their child with POMS coming to a scheduled visit at the site. The research staff completed the "phone script for recruitment" to inform the potential participants about the study (see Appendix K for phone script for recruitment). In addition, potential participants and their children or adolescents with POMS who had an upcoming appointment but have not been previously seen at CPODD were screened for eligibility by the researcher prior to their upcoming

appointment as well. These potential participants were given a flyer during their visit and instructed to contact the researcher for further information about the study if interested.

After the potential participants expressed interest in participating and if the participants met eligibility for the study, the researcher obtained informed consent and authorization (see Appendix L for IRB approval for consent and authorization). In this process, all potential participants were informed of the following: 1) the purpose of the study, 2) how the study would be conducted, 3) how to contact the primary investigator, 4) the benefits of the study, 5) risk of participating, 6) assurance of confidentiality, 7) the sharing of results, 8) the convenience of conducting the study for the participant 9) incentives for participation and 10) information about a potential follow-up (Polit & Beck, 2012). There was a 24-hour waiting period between the time the potential participant was informed about the study and written informed consent was obtained for the participants conducting face-to-face interviews and or verbal consent for those participants conducting interviews by phone or via video conferencing.

Data Collection

After the waiting period, once an individual agreed to participate in the study and provided written consent, the data collection process took place at a scheduled time convenient for both the researcher and the participant. The data collection process consisted of two components: a demographic form and the interview. The participants had the option of conducting the interview with the researcher either in person, via video-conferencing using GoToMeeting™ or by phone (Lo Iacono, Symonds & Brown, 2016). GoToMeetings™ is a web-hosed service for on-line meeting and video conferencing that

enables the users to meet with other computer users in real time. Video conferencing can be used to conduct a session from computer to computer or from computer to phone via the internet. Internet based methods of communication are becoming increasingly important and influential as they offer new opportunities for researchers to capture participants that may have not been feasible to study in sociological research (Lo Iacono, Symonds & Brown, 2016). Likewise, the demographic form could be completed in person or it could be mailed to the participants prior to the interview for completion. The collection of demographic data provided background information regarding the participants, the child with POMS, and other adults and children living in the household and offered insight into family factors influencing adjustment and adaptation in families of children diagnosed with POMS (see Appendix M for socio-demographic form).

For a qualitative study of this nature, in-depth, semi-structured interviews were the primary process of data collection so that rich and in-depth descriptions of the phenomenon could be captured (Creswell, 2013). Each audiotaped interview conducted between the individual participant and the researcher lasted 60 to 90 minutes. The researcher developed an interview script and guide that contained open-ended questions with a series of pre-established probes to ensure that all the participants were asked the same questions (See Appendix N for the interview script). If the participant agreed to a phone interview, verbal consent was obtained over the phone and documented in the transcription and the interview was conducted immediately following the consent (See Appendix P for the verbal consent and phone interview script).

The in-person interviews were conducted in the clinic or center's office in a private room free of noise and traffic. If the participant agreed to a phone interview or

video conferencing, then the researcher would talk with the participant about scheduling a convenient day and time for the interview in a quiet area in the participant's home that will ensure privacy without interruption. The potential participant was reminded of the interview one week prior to the scheduled time and date of the interview using the appointment reminder script (see Appendix Q for the appointment reminder script).

Data collection began with ten participants and as innovative ideas continued to emerge the researcher decided to expand the inclusion criteria to recruit additional participants to ensure that saturation was reached and that the data obtained was rich and descriptive. Once permission and support was obtained from the adult neurologist regarding the recruitment of participants who were previously treated at the CPODD but who had now transitioned to the UAB Adult MS Center, the researcher submitted an amendment to the original IRB approval. The inclusion criteria were expanded to 1) include those caregivers of children diagnosed with POMS and receiving care in the last two years and 2) those caregivers of children who were ≤ 22 years of age but who had transitioned the UAB Adult MS center for ongoing care. An amendment to the original IRB approval was then filed and approved (see Appendix R for amendment to IRB approval). With descriptive inquiry, a smaller sample is justified due to the in-depth and detailed nature of the interviews (Sandelowski, 2010). Qualitative research is based on the informational needs of the study with data saturation as the guiding principle as there are no fixed rules for sample size in this type of research inquiry (Sandelowski, 2010). Data saturation is achieved when data collected from interviews no longer yield new information and becomes redundant (Polit & Beck, 2012).

Over a 14-week period of enrollment, 20 potential participants, who met the prior eligibility requirements, contacted the researcher about the study. Although data saturation was reached at the 15th participant, the additional five participants were interviewed to ensure that there was rich and descriptive data for this study. This enrollment number met the typical sample size for qualitative descriptive design studies that range from three to 20 participants (Magilvy & Thomas, 2009). All interviews were recorded using the appropriate device based on the nature of the interview. For the in-person interview, the researcher used a digital voice recorder and the recording was saved as a MP3 (audio only) file. For the interviews conducted via phone, a phone to pc audio adapter was used along with the digital voice recorder to record the interview and then saved using a MP3 (audio only) file. For the interviews conducted via video conferencing, the interviews were recorded and saved as a MP4 (video and audio) file. This enabled the researcher to view the emotional responses as well as listen to the participants after the interview was complete.

A certified transcriptionist transcribed all interviews verbatim. The researcher checked each transcript for accuracy by comparing the transcript and the audio recording. In addition to the transcribed interviews, the researcher completed field notes to capture any insight, personal emotion, the demeanor of the participant, or the context of the situation that may further enhance the description of the phenomenon (Shenton, 2004). These notes were documented within 24 hours so that the essence of their importance is not lost over time.

Data Analysis

After each interview was transcribed, the data from each transcription was analyzed. Data collection occurred concurrently with data analysis. Thematic approach was used to analyze the data. Nursing researchers often use qualitative thematic analysis as an approach to data analysis in qualitative descriptive research. Thematic analysis is defined as the identification, analysis and reporting of themes within data that will provide rich and detailed, yet complex, accounts of the data with common threads across multiple sets of interviews (Vaismoradi, Turunen, & Bondas, 2013). Thematic analysis is the best approach to capture the very essence of a phenomenon that affects a family and their adaptation to a process that not only affects both the child with the diagnosis but the entire family (Ganong & Coleman, 2014). These families come from various backgrounds, may be in very different phases of acceptance, and have different skills within their family structure that will enable various levels of coping and adaptation. Thematic analysis allows these differences to be captured and allows commonalities to be identified.

(Braun & Clarke, 2014) described the following steps as the processes of thematic data analysis: 1) familiarizing oneself with the data; 2) generating initial codes; 3) searching for themes; 4) reviewing themes; 5) defining and naming themes; and 6) producing the report. In the first step, *familiarizing with the data,* the researcher reads and rereads the data and notes his or her initial ideas (Braun & Clarke, 2014). The second step, *generating initial codes,* entails coding of interesting features within the entire data set in a systematic manner. NVivo 11, a computerized software used in qualitative analysis for data storage and organization, was used to organize data so that it

can be coded or "bracketed" for significant statements to achieve horizontalization (Braun & Clarke, 2014; Leech & Onwuegbuzie, 2011). Horizontalization refers to the process wherein the researchers lists each of the relevant quotes of the studied topic and gives them equal value with regard to the expression of the group (Moustakas, 1994). With this step, all relevant data are gathered and placed under the potential theme. In the fourth step, *reviewing themes,* the researcher checks to see if the themes work in relation to the coded extracts and generates a thematic map of the developed codes (Braun & Clarke, 2014). *Defining and naming themes,* which is the next step in analysis, consists of the refinement and ongoing analysis of each theme and the overall story that the analysis will tell (Braun & Clarke, 2014). In this step, definitions and names of themes are generated. The last step in thematic analysis is *producing the report,* in which the researcher has a final opportunity for analysis (Braun & Clarke, 2014). This step involves the researcher selecting vivid examples from the selected extracts and relating these themes back to the research question and the literature to produce a report rich with details.

Information from the demographic data form were analyzed using SPSS software. Descriptive statistics of the demographic data were computed for all participants to develop descriptive analysis pertinent to the proposed study. Demographic data was also used to provide a detailed description of the participants and their families from a socio-economic view to validate the experiences of the families with the participants' interview to ensure that the research questions were fully explored.

Trustworthiness and Integrity of the Research

Quality assurance was guided by the trustworthiness of the study. Lincoln and Guba's framework of quality criteria will guide the establishment of trustworthiness of the study. Lincoln's and Guba's framework suggests four criteria for developing trustworthiness: credibility, dependability, confirmability and transferability (Lincoln & Guba , 1985; Polit & Beck, 2012; Shenton, 2004).

Credibility

Credibility or the believability of the study is similar to internal validity in quantitative research. This study, the researcher achieved credibility through prolonged engagement, persistent observation and triangulation, and member checking. Prolonged engagement involves investing sufficient time to meet the participants and establish trust, learn the participant's culture, and confirm inaccurate information from the researcher or the participants (De Chesnay, 2015). The researcher developed the interview guide and the demographic form and pilot tested these instruments with two caregivers of children diagnosed with POMS who were not eligible for the study because their children were more than 22 years of age. These individuals were given the interview guide and the socio-demographic questionnaire to review to ensure that the questions were worded in a way that was understandable to the reader. The researcher used feedback from these individuals to make any changes to the interview guide prior to the study onset.

Persistent observation during the interviews ensured that appropriate characteristics of the participants were identified relevant to the phenomenon under study so that premature closure of the study would be avoided (De Chesnay, 2015). The researcher gathered detailed demographic data and used open-ended questions to derive a

rich description of the phenomenon. The researcher used member checking, a technique during the interview process in which the researcher validates information with the participants by discussing and clarifying their answers as the interview is being conducted. (Shenton, 2004).

Dependability

Dependability or consistency of the study goes hand in hand with credibility. In the case of this study, the researcher selected an independent auditor who was unbiased to the study. This person utilized audit notes, transcripts, and data collection instruments during the development phase so that data could be tested and recorded and necessary changes could be made in a systematic manner (Shenton, 2004).

Confirmability

Confirmability or neutrality of a study is the way in which a researcher establishes the findings based on the subjects and their experience of the phenomenon and not on the biases, motivations, interests and perspectives of the researcher (Shenton, 2004). In this study, the researcher maintained a daily journal of activities, insights, and decisions about the study and its methods.

Transferability

Transferability, the final criteria for trustworthiness, refers to the applicability of the study to other contexts or with other subjects (Shenton, 2004). Transferability is difficult to achieve in qualitative studies, because of the small number of participants which makes the findings difficult to generalize outside of the research context (Shenton, 2004). In this study, the researcher supported the highest level of transferability by

providing a rich, thick description that produced the most information and allowed for transferability by others.

Protection of Human Subjects

As with all research studies, the maintenance of ethical conduct throughout the proposed study is of utmost importance. Ethical conduct includes obtaining institutional review board (IRB) approval, the protection of human rights, confidentiality, informed consent, beneficence, respect for human dignity and privacy. These aspects will be discussed in the order mentioned.

Institutional Review Board

Because of the possibility of a biased self-evaluation in research, research should be subjected to external review. The IRB reviewed the research plan to ensure that risks to participants were minimized, that informed consent was sought, and that monitoring of research was in place to ensure the safety of all the participants (Polit & Beck, 2012). IRB approval was sought prior to beginning the research to ensure that the plan met federal, local, and institutional requirements for ethically sound research (Buchanan & Huang, 2012).

Protection of Human Rights

The protection of human rights was maintained at all times; from the planning phase of the study to the enrollment and recruitment period, throughout data collection, analysis and dissemination of results as well as after the research was formally completed (Polit & Beck, 2012). Prior to the start of the study, the researcher completed a risk/benefit assessment and formal IRB approval was obtained.

Confidentiality

Because of the researcher's status as the healthcare provider of the participant's children, anonymity was difficult to maintain within the context of the interviews. However, anonymity of study participants outside of the research were maintained by the researcher to ensure the safest means of protecting the participants 'privacy and confidentiality (Polit & Beck, 2012). Great care was given by the researcher to not discuss any of the study finding with the other healthcare providers or staff involved in the care of the participants and their family. The identity of the participants were concealed through the distribution of pre-coded questionnaires that did not request information that could identify the participant. Additionally, study findings were reported in aggregate, which helps maintain study participants' confidentiality. The researcher put safeguards in place to ensure that confidentiality was maintained by ensuring that individual information was not publically reported or made accessible to anyone. In addition, the interviews were conducted in a private room free of traffic in order to prevent the identification of the participants.

In addition, the researcher ensured that a breach of confidentiality did not occur by keeping the transcribed interviews on a password-protected computer to prevent unauthorized individuals from accessing the data. In addition, the data was de-identified to maintain confidentiality and each participant was assigned an identification number. Names, social security numbers, or any other personal identification potentially linking participants to this study were kept separate from the data in a locked file cabinet in the researcher's office.

If the participant chose to complete their interview via phone and completed the socio-demographic form by mail, confidentiality was maintained for those participants as well. The form contained the participant's identification number only and did not contain any identifying data. A self-addressed stamped envelope was sent along with the forms to the participant for return of the form. There was no identifying information on the envelope to indicate its contents or the participant's identity.

Informed Consent

A third component of ethical research is informed consent. Informed consent is defined as providing sufficient information to potential participants that they have the ability to consent or decline to voluntary participation (Cahana & Hurst, 2008). The consent form was written at an 8th grade reading level and included the following information: Study goals and procedures, expected time commitment including potential follow-up, research sponsorship, and potential benefits and risks of participating (Polit & Beck, 2012). In addition, the consent form included information about compensation, maintaining confidentiality, the right to withdraw or withhold information without penalty, and the contact information of the researcher and the IRB. To ensure that the potential participant understood the research to which he or she was consenting, the researcher read the consent form aloud and allow for questions and clarifications prior to obtaining written or verbal consent from each participant. Written consent served as documentation that participation was voluntary (Cahana & Hurst, 2008). A copy of the consent form was given to the participant for their record (See Appendix O for consent and authorization form). Verbal consent was obtained if the participant chooses to participate in the research by phone or video conferencing. The consent was documented

as part of the transcribed interview. Verbal consent was documented on the consent form by the researcher and a copy of the consent was mailed to the participant for their records.

Beneficence

The Belmont Report (1979) established three principles for the protection of human subjects; beneficence, respect for human dignity, and justice. Beneficence requires the researcher to minimize harm while maximizing benefits to the participant (Polit & Beck, 2012; Townsend, Cox, & Li, 2010). In this study, minimal risk of harm was imposed based on the qualitative nature of the research methodology. In a qualitative study of this nature, psychological distress is the greatest risk because participants have their feelings, fears and weaknesses exposed during the interview process (Polit & Beck, 2012). In order to minimize the risk, the researcher maintained consistent monitoring during each interview to recognize the participant's emotional reaction, provided frequent breaks during the interview when they participant appears distressed in person, over the phone or by video conferencing and provided the participant with an opportunity to stop, regroup or continue the interview (Draucker, Martsolf, & Poole, 2009). The researcher made a follow-up phone call within 24 hours using the follow-up phone script with any participant who experienced any form of psychological distress using the follow-up phone script (See Appendix S for Follow-Up Phone Script). A service at Children's of Alabama was put in place for additional counseling for those participants who remained in distress beyond the 24-hour period when the researched placed the follow-up phone call. This service was available by phone for those participants who conducted phone interviews as well. None of the participants required this service. The researcher was

prepared to deal with this distress in a professional and empathetic manner with the ability to continue in the research if the participant was willing. The researcher's sensitivity to this possibility was essential but the maintenance of probing was necessary to aid the researcher in asking questions that would lead to the meaningful results. Along with beneficence, respect for human dignity and justice are established by the Belmont Report as well (Polit & Beck, 2012).

Respect for Human Dignity

Respect for human dignity deals with the right to self-determination and the right to full disclosure. Self-determination is the right of potential participants to decide whether to participate without risk of prejudice or penalty (Polit & Beck, 2012). The researcher conducting this study ensured when obtaining informed consent that the potential participants understood the rights of self-determination. Potential participants were assured that they could ask questions about any aspect of the research study, refuse to provide information, and withdraw from the study at any time. Prior to the interview, the researcher informed potential participants that their beliefs, lifestyles, and habits would be respected in line with their culture or background. The right of full disclosure, another aspect of informed consent, is the nature in which the researcher fully describes the study to the potential participants. The researcher fully disclosed all aspects of the research, but took care in disclosing research questions that may create participant bias and recruitment bias.

Justice includes the participants' right to privacy and fair treatment (Townsend et al., 2010). The researcher maintained the right to privacy by conducting the interviews in a private office with a closed door to prevent the identity of the participant from being

exposed. Research conducted over the phone or by video conferencing was handled in a likewise manner. In addition, confidentiality, which maintains the right to privacy, was maintained before, during and after the research was conducted.

Implications for Being the Researcher and the Provider

Being the researcher as well as the participants' healthcare provider brought a unique set of challenges for the researcher. Two ethical principles, freedom of coercion and the right to fair treatment will be discussed regarding this unique role. In addition, other aspects of this role were addressed to ensure that ethically sound research was conducted.

Freedom from coercion, which falls under respect for human dignity, was of great concern when the researcher is also the participant's healthcare provider. Coercion involves threats of penalty if an individual decides not to participate or can involve the excessive reward if an individual agrees to participate (Polit & Beck, 2012). In this study, the researcher ensured that coercion did not occur by informing the potential participants that their child's treatment would not be affected by their decision to participate or not to participate. To avoid coercion due to financial compensation for participation in the study, the researcher gave all participants a pre-determined incentive of $25 required for their participation in the study. An incentive was offered only to compensate participants for their time.

To prevent coercion in the recruitment stage of the study, the researcher will email or mail recruitment flyers to all potential participants who have been pre-screened and ensure that contact information of the researcher is included on the flyer. The flyer will specifically state that if a potential participant would like to enroll in the study, they

should contact the researcher prior to their next appointment to work out a convenient time that the study will be conducted. Once the potential participants contact the researcher and inquire about enrollment into the study, then the researcher can ask the potential recruits if they would like to enroll in the study. At a routine visit, after routine examination and care, families will be engaged by the nursing staff and asked if they received the recruitment flyer, have any questions regarding the study, and inquire if they want to participate. For the potential recruits who state that they did not receive the recruitment flyer for any reason, the staff will distribute the flyer and the potential recruits asked to call the researcher if they would like to participate at another time. For the potential recruits that are traveling from out of town, a phone call may be made a few days prior to their appointment research staff to ask if they received the flyer and if they would like to participate after or before their upcoming clinic appointment to make better efficiency of time and resources related to travel. This will ensure that all potential recruits have the opportunity to participate in case they did not receive the flyer via mail or email for any reasons and will decrease the risk of coercion as well.

In addition to freedom of coercion, the researcher addressed the right to fair treatment of potential participants. The researcher ensured that the participants understood that they had the right to withdraw from the study at any time without prejudice. Each participant was assured that the care their child received from the researcher as a healthcare provider would not be compromised if they decided not to participate or decided to discontinue their participation in the study after being previously enrolled. In addition, the researcher was careful to choose a well-rounded group of participants and not prey on those individuals of less socioeconomic standards so that the

study would be unbiased. In addition to freedom of coercion, the researcher will address the right to fair treatment of potential recruits. The researcher will be careful to choose a well-rounded group of participants and not prey on those individuals of less socioeconomic standards so the study will be unbiased. The researcher also ensured that the potential participants understood that they have the right to withdraw from the study at any time without prejudice in the care that they received from the healthcare providing who is conducting the research. Prior to the interview taking place, the researcher informed that participants that their beliefs, lifestyles and habits will be respected despite their culture or background.

Another issue emphasized with regard to the healthcare provider functioning as the researcher included the confidentiality of the information revealed in the interview by caregivers of patients for whom the researcher provided care. The researcher was careful not to disclose any information discussed by the caregiver to the participant's family, including the child with POMS, at any time after the interview was complete.

Summary

A sound research proposal is well planned. This chapter outlines the research study that explored how factors influence the adaptation of families to a diagnosis of pediatric MS. This chapter outlined the research design and methodology chosen by the researcher based on the desire to answer the research question. The sampling strategy, recruitment plan, setting and data collection and analysis were presented in a detailed manner so that another researcher could replicate the study with ease. The chapter

concluded with a discussion of the strategies that were utilized to ensure validity and reliability while maintaining the protection of human subjects.

CHAPTER 4

FINDINGS

The purpose of this study was to explore the caregiver perspectives of how family factors influence adaptation in families of children with (POMS) seen at a southeastern specialty center. In this chapter, the results of this qualitative descriptive study are reported by sections. The first section is a description of the characteristics of the sample, which includes the sociodemographic data as well as the characteristics of the other family members that live in the household with the caregivers including the child diagnosed with POMS. The second section presents seven themes with the corresponding sub-themes. Six of the seven themes have corresponding sub-themes. The demographic data were analyzed using IBM SPSS Statistics Version 23 (IBM Corporation, 2015). The qualitative data included 20 individual interviews that were analyzed using thematic data analysis supported by NVIVO 11 Pro Version (QRS International Pty Ltd., 2015).

Sociodemographic Data

Study Participants

Sixty-seven primary caregivers of children who were treated with a disease modifying therapy for multiple sclerosis (MS) were screened to participate in the study from the Center for Pediatric Onset Demyelinating Disease (CPODD) database. Fifteen of the 67 potential caregivers were excluded because their child was older than the age of 22.

Three of the 67 potential caregivers had children that did not fulfill revised 2010 McDonald Criteria and the 2013 International Pediatric Multiple Sclerosis Study Group criteria for the diagnosis of POMS. Twenty-seven of the 67 potential caregivers were excluded because they had not been seen in the last two years at CPODD or the Adult MS center at UAB and one potential caregiver was ineligible because the child with POMS had been diagnosed less than 6 months prior to the beginning of the study. Twenty-six potential caregivers were sent a letter of support and information from Dr. Jayne Ness or Dr. Khurram Bashir along with a flyer that included the study's purpose, eligibility of the caregivers, a description of the study that entailed and the rights, benefits, and risks of participating as well the plan for maintenance of confidentiality. Dr. Jayne Ness is the pediatric neurologist at the CPODD and Dr. Khurram Bashir is the adult neurologist at the UAB Adult center. The flyer and letter both contained contact information for the researcher if the caregiver wanted more information about the study or wanted to enroll in the study. Twenty of the 26 eligible caregivers enrolled in the study.

All 20 caregivers completed the socio-demographic data form in its entirety and participated in an individual interview with the researcher as well. The caregivers' self-identified the following on the socio-demographic form: age, relationship to the child with POMS, education, marital status, employments status, health related issues, annual household income, race, and religious preference. The demographic data regarding the caregivers are listed in Table 2.

Table 2

Socio-demographics of the Caregivers (N=20)

Characteristic	n	%
Biological mother	19	95
Maternal aunt	1	5
Age Range		
<30	1	*5*
31-40	4	20
41-50	10	50
51-55	5	25
Race		
White	11	55
Black/African American	8	40
Multi-racial	1	5
Marital Status		
Married/domestic partnership	14	70
Single	4	20
Divorced	2	10
Religion		
Baptist	10	50
Pentecostal/Holiness	3	15
Catholic	2	10
Non-denominational	2	10
Orthodox	1	5
Protestant	1	5
Christian	1	5
Education		
High school diploma/GED	3	15
Some college/no degree	7	35
Associate's degree	2	10
Bachelor's degree	5	25
Technical/trade/vocational	2	10
Master's degree	1	5
Annual Household Income		
Under $10,000	2	10
$10,000-$19,000	3	15
$20,000-$29,000	0	0
$30,000-$39,000	1	5
$40,000-$49,000	5	25
$50,000-$74,000	5	25
$75,000-$99,000	1	5
$100,000-$150,000	3	15
Employment Status		
Employed for wages	12	60
Self-employed	1	5
Student	1	5
Homemaker	2	10
Unable to work	4	20

All the caregivers were female. Nineteen of the 20 caregivers identified themselves as biological mothers of children with POMS (n=19; 95%). One self-identified as being a maternal aunt to the child with POMS (n=1; 5%). The caregivers ranged in age from 28 to 55 years with a mean age of 44. A little over half of the caregivers were White (n=11; 55%), 40% were black (n=8) and one caregiver identified herself as multi-racial (n=1; 5%). Over two-thirds of the caregivers identified themselves as being married (n=14; 70%). Half of the caregivers identified their religious preference as Baptist. Eighty-five percent of the caregivers identified themselves as having at least some college, technical/vocational trade or a Master's degree.

Fifty percent of the caregivers reported an annual household income between $40,000 and $74,000 with a mean household income of $49,000. More than half of the caregivers identified themselves as employed for wages (n=12; 60%). Two-thirds of the caregivers identified themselves as being under the care of a healthcare provider for a chronic health care issue (n=13; 65%). Table 3 lists all the health care issues reported by the caregivers. Some of the more serious chronic conditions that required ongoing treatment included neurofibromatosis Type 2 tumors, dysautonomia, diabetes, multiple sclerosis, rheumatoid arthritis, and Sjogrens.

Children with POMS

Caregivers also reported socio-demographic data for the child with POMS, which included age at time of interview, sex, age at diagnosis, and number of years the child experienced symptoms prior to diagnosis. Caregivers also reported the information regarding the care of the child with POMS: (1) type of provider delivering routine MS care and treatment; (2) frequency of common symptoms; (3) use of disease modifying

therapies (DMTs); (4) number of flare-ups during the previous two years that required additional services; (5) number of permanent symptoms children with POMS experienced

Table 3

Caregivers' Healthcare Issues

Healthcare Issues	n
Anxiety	3
Carpel Tunnel	1
Chronic back pain	1
Diabetes	1
Dysautonomia	1
Fibromyalgia	1
Hearing Loss	1
High Cholesterol	1
Hypertension	4
Hypothyroidism	2
Leukopenia	1
Migraines	1
Multiple Sclerosis	1
Neurofibromatosis Type 2 tumors	1
Obsessive compulsive disorder	1
Osteoarthritis	2
Panic Attacks	1
Post Cancer follow-up	1
Post-traumatic stress disorder	1
Rheumatoid Arthritis	1
Sjogrens	1
Sleep apnea	1
Supraventricular tachycardia	1

Note. N=13 caregivers

since the diagnosis and (6) miles the family traveled to CPODD. The CPODD is the only center in the southeast that specializes in POMS; consequently, many caregivers and children with POMS must travel a great distance to receive routine care. The expenses of travel along with routine care and treatment of a child with POMS can have an impact on how caregivers and families of children with POMS adapt and adjust after the diagnosis is made. Table 4 lists the number of miles that families travel for routine MS care.

The caregivers reported that the ages of the children with POMS at the time of the

interview ranged from age seven to 22 years. Fifty percent (n=10) of the children with POMS were between 16 and 17 years of age. More than two-thirds of the children with POMS were females (n=14; 70%). Half were reported as white (n=10) while 40% were reported as black (n=8) with the remaining reported as being multi-racial (n=2; 10%).

Table 4

Number of Miles Families Travel for Routine MS Care

Mileage One-Way	N	%
<50	4	20
71-100	3	15
101-200	10	50
>200	3	15

The caregivers also reported data regarding the age at diagnosis of the children and the number of years the children with POMS experienced symptoms prior to the diagnosis. The youngest children ages at diagnosis were four and eight years of age. The mean age at diagnosis was 13.45 years. Sixty-five percent of the children with POMS (n=13) were diagnosed between 14 and 17 years of age. Seventy percent of the children with POMS (n=14) experienced symptoms for less than one year prior to being diagnosed with POMS; 20% (n=4) experienced symptoms between one and two years prior to diagnosis and 10% (n=2) experienced symptoms between four and five years prior to the diagnosis.

Nine (55%) caregivers reported that their child with POMS received routine care at CPODD for primary MS care every three to six months. Four (36.3%) caregivers reported that their child with POMS had transitioned to an adult neurologist for ongoing care and were seen every six months to a year for routine care. Of those children with POMS age 18 or older (N=7), two-thirds (n=5; 71%) were being treated by an adult

neurologist and one-third (n= 2; 28% were treated by a pediatric neurologist or a pediatrician. Of those children who do not seek routine MS care at CPODD, 71% (n=5) were seen at CPODD at least once a year for annual consultation and recommendations.

The caregivers in the study provided information on the disease modifying therapy (DMT) that their child was prescribed at the time of the interview. This information is displayed in Table 5. Less than half of the caregivers reported that their child was using one of the first line injectable treatments (i.e. Rebif®, Copaxone®, or Plegridy®) for MS (n=7; 45%) while less than a third of the caregivers reported that their child was using one of the oral medications, Gilenya®, as treatment for MS (n=6; 30%). One of the caregivers reported that her child with POMS was enrolled in a study that consisted of an oral medication and an injection. The remaining one-third were treated with second line agents such as Rituximab®, Tysabri® and Ocrelizamab® (n=6; 40%). More of the children of the caregivers in this study were not taking first line DMTs that are typically given to POMS patients. Indications for intravenous medications and oral medications in POMS patients are usually due to increased disease activity or non-adherence with the method of delivery of first line DMTs (subcutaneous injection).

Table 5

Disease Modifying Therapy (DMT) Used by Child with POMS

DMT	N	%
Clinical Trial Drug	1	5
Copaxone®	4	20
Gilenya®	6	30
Ocrelizamab®	1	5
Plegridy®	2	10
Rebif®	1	15
Rituximab®	3	15
Tysabri®	2	10

The caregivers reported symptoms that their child with POMS experienced that require medications. They also reported the number of flare-ups experienced by their child with POMS in the first two years and if those flare-ups required additional services such as outpatient physical or occupational therapy, speech therapy, outpatient infusion of steroids or immunoglobulins, an overnight observation in a hospital, a hospital admission or an admission that required extensive rehabilitation. In addition, the caregivers reported if the child with POMS experienced any permanent symptoms of MS such as anxiety, blindness, depression, heat or cold sensitivity, impaired attention and concentration, impaired use of upper extremities, loss of bladder or bowel control, alterations in mood, and the need for a wheel chair. This information was gathered to indicate the extent to which families may need to be involved in the care of the child with MS.

Half of the caregivers reported that their children were being treated with medication for MS related symptoms (n=10; 50%). Table 6 lists the symptoms and the number of children being treated for the symptom. Of those reported by caregivers as having symptoms that need to be treated, two of the children were being treated for two concurrent symptoms (20%) and six were reported as being treated for three concurrent symptoms (30%). The most common symptoms treated were neuropathic pain, depression, and anxiety followed by attention/concentration.

A flare-up is an indicator of disease activity. After the diagnosis, most POM patients have very active disease as they transition to a DMT and their immune system begins to adjust to the changes that the brain and body are experiencing. The caregivers

reported the number of flare-ups experienced in the first two years by the child with POMS that required additional services. Only one caregiver reported that her child with POMS did not have a flare-up within the first two years of diagnosis. Nearly all

Table 6

Symptoms Treated by Medication for Children with POMS (n=10)

Symptoms	n
Anxiety	4
Attention/Concentration	3
Depression	4
Fatigue	2
Headache	1
Mood	2
Neuropathic Pain	4
Sleep Disturbance	2
Tremor	1

caregivers (n=19; 95%) reported that the child with POMS required outpatient infusion of Solumedrol® at least one time during the first two years. Nearly half of the children with POMS required physical therapy, occupational, speech or water therapy during the first two years due to a flare-up. Only a small percentage (n=2; 10.5%) required an overnight stay at the hospital due to a flare-up. More than two-thirds (n=13; 68.4%) required a two to five-day stay at hospital and nearly a third (n=5; 26.3%) required a one to two week stay at the hospital. None of the caregivers reported a flare-up that required admission for rehabilitation. Caregivers were not asked if more than one service was required during each flare-up.

In addition to the first two years, the caregivers reported the total number of flare-ups that the child with POMS experienced that required at least a doctor's examination, or any other professional treatment, care coordination or hospitalization. This is an indicator of ongoing disease activity since their diagnosis. All the caregivers reported at least one

flare-up since being diagnosed. More than two-thirds of the children with POMs were reported as having three or four flare-ups since the diagnosis (n=13; 65%); a fourth of the children had at least one flare since their diagnosis (n=5; 25%) and one child (5%) had six flare-ups since his/her diagnosis that required some form of treatment

Permanent symptoms are the residual effects of MS as the result of the damage caused by a flare-up. These symptoms do not go away and may or may not be treated with medications, but may pose yet another issue for families of children with POMS. More than three-fourths of the children with POMS were reported as experiencing permanent symptoms (n=14; 70%). Table 7 lists the permanent symptoms experienced and the number of children with POMS reported to experience the symptoms at the time of the interview. The most common permanent symptom reported were heat/cold intolerance, followed by impaired attention/concentration, anxiety and mood alterations. Of those that reported permanent symptoms, two-thirds of those children with POMS were reported to have at least one or two co-existing permanent symptoms (n=10; 71%); one caregiver reported their child as having five co-existing permanent symptoms (n=1; 7%) and one caregiver reported their child as having nine co-existing permanent symptoms (n=1; 7%).

In addition to the caregivers providing socio-demographic data about themselves and the child with POMS, self-reported information was provided regarding the other adults (> 18 years of age) and children (≤ 18) living in the household. Socio-demographic data regarding the other adults living in the household included the following: age, relationship to caregivers, employment status and health related issues for which they are seeking a healthcare provider. The information regarding the other adults

and children living in the household provided insight into the make-up of the household. It also provided insight into issues that families may be facing other than those issues regarding themselves or those regarding the child with POMS.

Table 7

Permanent Symptoms Experienced by POMS Patients (N=14)

Permanent Symptom	n
Anxiety	6
Blindness/severe visual dysfunction	1
Depression	4
Heat/Cold Sensitivity	12
Impaired attention/concentration	6
Impaired use of upper extremities	1
Loss of bladder/bowel control	1
Mood alterations	5
Need for wheelchair/walker	1
Severe cognitive dysfunction	1

Other Adults Living in the Household

Three-fourths of the caregivers reported having additional adults living in the household (n=15; 75%). Table 8 displays the socio-demographic data regarding the other adults living in the household. The ages of the adults reported by the caregivers ranged in age from 19 to 64. Fifteen caregivers reported 23 adults living among all of them. More than half of the adults (N=15) living with the caregivers were spouses (n=13; 56%) and a little over one-fourth of the other adults living in the household were reported as children of the caregivers (n=6; 26.1%). One of the caregivers reported living in the household with both her parents (n=2; 8.7%) and two caregivers reported living in the household with their sibling (n=2; 8.7%).

All the spouses living in the household were employed for wages. Forty percent (n=4) of the other adults living in the household were employed for wages, 40% (n=4) were full-time students, and 20% (n=2) were unable to work. The caregivers reported

that two-thirds of the spouses living in the household (n=8; 62%) were under the care of a healthcare provider; three-fourths of those spouses (n=6; 75%) having co-existing conditions for which they are under the care of a healthcare provider.

Table 8

Socio-demographics of the Adults Living in the Household

Characteristic	N	%
Age of Adults Living in the Household (N=23)		
19-29	7	37.3
30-39	1	4.3
40-49	9	39.1
50-59	4	17.3
60-69	2	0.8
Employment Status		
Spouses (n=13)		
Employed for Wages	12	92.3
Self-Employed	1	7.7
Other Adults Living in the Household (n=10)		
Employed for Wages	4	40
Unable to Work	2	20
Students	4	40

Thirty percent of the other adults living in the household (n=3) were under the care of a healthcare provider; two out of three of those were identified as having co-existing conditions. The health conditions experienced by all adults living in the household are listed in Table 9. The two most common conditions were hypertension and diabetes. None of the adults who were children of the caregivers was as reported as having a condition to be managed by a healthcare provider.

Other Children Living in the Household

Socio-demographic data regarding the other children that were less than 18 years of age living in the household included the following: age, sex, relationship to the caregiver, and health related issues. As reiterated with the data collected on the other adults living in the household, healthcare related issues were reported to identify any insight into

Table 9

Healthcare Issues for Other Adults Living in the Household

Healthcare Issue	n
Arthritis	1
Diabetes	5
GERD	1
High Cholesterol	1
Hypertension	5
Irritable Bowel Syndrome	1
Seizures	1
Sleep Apnea	1
Thyroid Disease	1

N. Note N=11 other adults living in the household

issues that caregivers may be faced with when providing care for other children living in the household along with the child with POMS.

The age range of children living in the household was five months to 18 years of age. Nearly two-thirds of the reported children living in the household were males (n=9; 60%) and more than half were reported as being children of the caregiver (n=11; 73. 3%); two of the children were nieces of the caregiver (13.3%) and two of the children were grandchildren of the caregiver (13.3%). Four of the 15 children (26.7%) were reported as having a health condition that required the care of a healthcare provider and only one of the children was noted as having co-existing conditions.

There were a variety of conditions reported including chronic illnesses such as asthma, attention deficit disorder, cyclic neutropenia, post-traumatic stress disorder and Trisomy 21.

Themes with Corresponding Sub-Themes

The twenty interviews of primary caregivers of children with POMS were analyzed using Braun and Clark's six steps to thematic analysis (Braun & Clark, 2006).

The analysis resulted in seven major themes, which reflected the collective perceptions of primary caregivers of children on the adaptation of families of children with multiple sclerosis. The first theme, "stress and strain" was related to the identified demands on the family of the child diagnosed with POMS, both related and unrelated to the diagnosis. The second theme, "adjusting to the diagnosis" reflected the family's appraisal of the diagnosis and their perception of the impact on their lives. The third theme, "communication", reflected the way in which families' problem-solve to decrease levels of stress caused by the disease. The fourth theme, "coping with the diagnosis" reflected the identification of strategies and patterns to maintain the strength of a family. The fifth theme, "sources of strength" identified resources utilized by the family as well as their capabilities as individuals to manage stress and restore harmony within the family. The sixth theme, "achieving balance" identified how families achieve balance between the demands of the POMS and the needs of the family. The seventh and final theme, "the overall experience of the family", provided a summary of each family's experience from the perspective of the caregivers as a means of reflection and insight for the researcher.

Six of the seven themes all had sub-themes and are expanded in the following sections. The themes and sub-themes are illustrated by selected, direct quotes from the caregivers that represent their experiences regarding the adaptations as a family. To maintain anonymity and confidentiality of the caregivers, each of them were assigned a pseudonym. The pseudonym along with a description of the caregiver and their family structure is listed in Table 10.

Table 10. Pseudonyms of Caregivers

Caregiver	Pseudonym	Descriptive Characteristics of the Caregiver and the Family
1	Sue	44-year-old married mom with a child now age 16 diagnosed with POMS at 13 years of age living with 2 other biological children (age 9 and 19) in the household
2	Donna	28-year-old unmarried mom with a child now age 7 diagnosed with POMS at age 4 years of age living with 3 other adults in the household (2 parents and a younger sibling)
3	Joan	48-year-old married mom with a child now age 18 diagnosed with POMS at age 17 living with 2 other biological children in the household (age 14 and 19)
4	Tina	42-year-old married mom with a child now age 16 diagnosed with POMS at age 13 living with 1 other biological child (age 20)
5	Heather	40-year-old single mom with a child now age 22 diagnosed with POMS at age 15 living with 1 other biological child (age 11)
6	Katie	44-year-old divorced mom with a child now 18 diagnosed with POMS at 17 years of age living with 1 other biological child (age 11)
7	Carol	51-year-old married mom with a child now age 17 diagnosed with POMS at age 14 years of age living with 1 other biological child (age 20)
8	Denise	44-year-old single maternal aunt with a nephew now age 16 diagnosed with POMS at 15 years of age with no additional individuals living in the household
9	Sadie	38-year-old married mom with a child now age 18 diagnosed with POMS at 16 years of age living with 1 other biological child (age 12)
10	Morgan	39-year-old married mom with a child now age 16 diagnosed with POMS at age 15 without any other people living in the household
11	Sarah	35-year-old married mom with a child now age 16 diagnosed with POMS at age 11 living with 1 additional biological child (age 14)
12	Maria	55-year-old single mom with a child now age 16 diagnosed with POMS at age 14 with no additional biological children and or adults living in the household
13	Judy	45-year-old married mom with a child now age 21 diagnosed with POMS at age 16 with 1 additional biological child in the household (age 18)
14	Nicole	45-year-old married mom but now living in the home with her spouse but with her biological sister along with a with a child now age 15 diagnosed with POMS at age 14 with no additional biological children living in the household

15	Shannon	46-year-old married mom with a child now age 22 diagnosed with POMS at the age of 13 living with 1 additional biological child (age 20)
16	Daisy	51-year-old divorced mom with a child now age 15 diagnosed with POMS at the age of 14 living with 1 additional biological child (age 12)
17	Lillie	45-year-old married mom with a child now age 21 diagnosed with POMS at the age of 14 living with 2 maternal nieces (ages 14 and 17)
18	Dollie	53-year-old married mom with a child now age 22 diagnosed with POMS at the age of 14 living with no other biological children but two biological grandchildren (2 years and 5)
19	Lisa	53-year-old married mom with a child now age 19 diagnosed with POMS at the age of 13 living with 3 other biological children (ages 14, 17 and 23)
20	Karen	41-year-old married mom with a child now age 16 diagnosed with POMS at the age of 8 living with 1 other biological child (age 13)

Stress and Strain

The caregivers in the study were asked to discuss any form of stress or strain that existed prior to their child being diagnosed with POMS that they experienced as a family. Family stress and strain prior to the diagnosis would give an indication of the demands that existed with these families prior to the diagnosis of POMS that may be a source of unresolved strain on the family. The caregivers were also asked about their initial reaction to the diagnosis and the changes that took place after the diagnosis with the family. This would indicate other family demands that were created by the diagnosis of POMS.

The theme “stress and strain” resulted from the analysis of the interviews and consisted of three sub-themes: a) *stress and strain prior to the diagnosis, b) initial reaction to the diagnosis and c) changes after the diagnosis.*

Stress and Strain Prior to the Diagnosis

Family stress and strain prior to the diagnosis can affect how families perceive and view the initial and ongoing demands of the new diagnosis (McCubbin & McCubbin, 1983; McCubbin, McCubbin et al., 1995). The stress and strain can have an emotional, financial and/or physical impact on the members of the family and there may be multiple stressors that would attribute to the pile of demands on the family. Half of the caregivers identified that they had the typical stressors prior to the diagnosis. Donna, Lillie, and Dollie identified such things such as the loss of a job of a spouse, single-parent income, parental health issues, and separation between the child and parent as well child versus stepparent rivalry.

Yet some of the caregivers revealed atypical stressors experienced by their families. Karen revealed in her interview, "I had three very difficult miscarriages right before (child with POMS) was diagnosed". Denise, the maternal aunt of the child with POMS, explained her sibling's condition in her interview:

> His mother had been diagnosed with Hashimoto's disease…she was probably diagnosed maybe a year prior to (child with POMS) diagnosis. Um, so that puts a lot of strain because of the Hashimoto's, she has a lot of memory issues, and she's got a lot of fatigue...that's very stressful actually, 'cause she'll forget things, she forget appointments, she'll forget conversations. Very stressful.

One of the caregivers provides her personal encounter with being diagnosed with MS prior to her son's diagnosis with POMS:

> So, as you know I was diagnosed with MS six years ago. Six going on seven years ago. We were dealing with that. I mean, I'm stable but we were dealing with that.

> (Child with POMS) even though he knew I had it, he still wasn't registering that okay this is serious and he didn't, since at that time it was only affecting me, he really wasn't, you know how teenage boys, he wasn't looking at or being overly concerned about it. But me personally I was dealing with that on my level trying to be strong for him and not letting him see what was going on with me as much and still trying to keep everything as normal as can be.

Of those caregivers that identified those more serious stressors, most of them identified one or two stressors; a few of them had multiple stressors that put them at risk of having more difficulty with their perceptions of the diagnosis of a new chronic illness. One of caregivers was Sarah. In Sarah's interview, she talked about a combination of events that she was dealing with prior to the diagnosis of POMS:

> The separation between their biological father and the kids... 'cause we moved right before. We moved from Mississippi to Florida. They were in Mississippi this whole time. I wasn't with their father, but we were close. So if he could pick them up from school, they could have that time. So, when we moved, they had to transition to not seeing their dad as much and adapting to staying away from him. So that was a big strain on our family. Cause we had to deal with that too, the burden of it. . .And then we went through Hurricane Katrina, which kind of lingered on years because that one thing affected our life long term. Even now, dealing with some weather and just mentally. So that was some stuff that we went through that made a big difference too…Okay her brother (sibling of child with POMS) had cyclic neutropenia, which is very rare. Still to this day, they're trying to figure out what is it that's taking his neutrophil levels and his leukocytes. They

> don't really function and do what they're supposed to do. And also, he had been having fevers all the time, random. So, we actually moved to Nashville so he could see the doctor who discovered periodic fever syndrome. So, we found out that's what he was suffering from. So, we went from there and then we ended up getting a study with the NIH clinic. So, he's on protocol with them. So, we were dealing with all that stuff before we even knew what it was. Early on.

Initial Reaction to the Diagnosis

Stress and strain experienced by families not only included the family's prior experiences with unresolved strains, but it also included the family's initial reaction to the diagnosis of POMS. The caregivers discussed their own personal reactions, the reaction of their spouses as well as the reaction of the child diagnosed with POMS and their siblings as well. These findings are in the following section.

The news of an unexpected diagnosis like POMS can be very emotional for a family. The primary caregivers of children with POMS were asked to describe their personal and family's emotions when they first heard the words 'multiple sclerosis'. The caregivers described a wide range of reactions to being told that their child or their relative had multiple sclerosis.

Two of the caregivers recollected feelings of helplessness when they received the diagnosis. Dollie stated:

> Oh Lord… I was really stressed. I cried a lot. 'Cause all I knew about it was that, you know, people that I knew that had MS were in wheelchairs. And I felt helpless… 'Cause as momma, you want to be able to do something. And I couldn't do anything to help her. So it was really, really emotional for years…it

> was emotional…It was very emotional and very stressful, and yeah, I had a few meltdowns.

One of the other caregivers recalled her feelings of helplessness due to her own chronic medical condition. In the interview with Daisy, she stated,

> Upon receiving the diagnosis, it was very, very difficult. I have chronic pain and so the difficulty for me is as a mother not being able to care for my child. I don't like to be vulnerable. I don't like to weak. With the diagnosis that I have, sometimes it hurts to be touched. Sometimes my body aches and hurts really bad, but if she needs me to assist her, my biggest concern is that I won't be able to provide assistance that she need because of my own disability. That's what frightens me.

Two of the other caregivers expressed feelings of shock and disbelief in their interview. In Lisa's interview, she stated:

> I was very shocked, because it started out just as a sinus infection. We went for … (Child with POMS) got sick with a sinus infection, and we treated it with over the counter stuff like you would normally do. …It never got better, so we went in and that when things began to unravel.

Carol stated in her interview, "I was shocked, and I didn't expect that it would happen to her." Another parent, Sadie, discussed her shock then a flood of emotions followed later by fear. Sadie recalled, "Probably shock at first, and then like I said, once I started doing more research it was more of a…like a weight on your chest, like "What is this going to do?"

Judy and Denise expressed feelings of devastation. When Judy was asked how she felt after receiving the diagnosis, she stated, “Unbelief. Mad. Was it my fault? How could this be? …One of the most devastating times in our life.”

Tina, Heather and Katie expressed their fear of the unknown with the diagnosis as well as their concern for their child’s future and being a normal child. Donna and Carol expressed their feelings of pity for the child being diagnosed with POMS. Carol stated, “It’s just that there are plenty of children who are born in this world and she’s the one who got this type of disease.”

Karen, Donna, Sue and Carol expressed a combination of mixed emotions of fear, relief, sadness, denial, scared, devastation, helplessness, terror and confusion when they received the diagnosis. Karen stated the following:

> Relief. Because I hadn’t an answer for two years. We hadn’t been able to figure out what was going on. So, I think, definitely relief and devastation and fear. (We had) …a certain amount of relief, that we know what this was all about, and fear of the unknown.

Maria expressed the following:

> Receiving ... shock. Denial. Sadness. Fear. Fear of what may happen. Fear of the unknown because you don't know. When you have a child, you don’t have these expectations but you have this thing on your mind that your child is going to grow up and be prosperous and have a family and be healthy and just do better than you've done in your lifetime. When you get a diagnosis like that, you start to think will my child. How will my child be in 20 years? Will he be prosperous? Will he have a family? Will he be able to support himself? So, you don't know. But, you

> still hope that because you want the best for your child. But when you get a diagnosis like this with no cure, that whole frame of mind is just thrown off. You know?

Although most of the caregivers expressed negative feelings after receiving the diagnosis, one of the caregivers expressed a sense of empowerment. Sarah stated:

> At first, I was overwhelmed. I couldn't cry because it was like, "okay, now what are we gonna do to beat this?" So, it was just like a resistance to come back and say, "Okay, what can I do on my end?" So, it was like I opened up to more ideas and how we're gonna deal with this. So, my emotion was, let's get to work. You know, sad but let's get to work. No pity party, if you can think of it that way.

The caregivers also discussed their observations of their children's reaction to the diagnosis of POMS as well as the reactions of the siblings and the fathers of the children with POMS. Most of the caregivers stated that their children with POMS were too young to understand their diagnosis and were not able to explain their feelings about it. A few caregivers expressed their children's emotions in the interview. Lillie recalled her daughter's apprehensions regarding her MS diagnosis (diagnosed at 14 years of age):

> (Child with POMS) was very concerned, because she wanted to know how serious it could be, what could happen to her, you know, was she gonna die or things of that nature, but I think my family was more concerned, because a lot of family members know exactly what it would do, unlike me.

Some caregivers elaborated on their observations of the feelings and reactions of their spouses. In Sadie's interview, she shared that her husband was quiet in the first few days and weeks after the diagnosis. Later, she stated that they discussed their concerns

and the need to rely on their faith. Some of the caregivers perceived that their spouses had unexpected feelings of defeat and loss of control and reacted in anger. The caregivers reported that these feelings and emotions were temporary; however, some lasted for years after the initial diagnosis. Shannon reported, “My husband felt defeated. He said he couldn’t protect her, like you know how you protect from the boys when she dates.” Tina stated the following about her spouse:

> My husband was the only one that had the issue (laughs). He had the issue. He was like, Oh my God. Like, Oh my God, I think he was depressed for a good month or so. I'm like, Dude, snap the hell out of it. You're getting on my nerves. She's gonna be fine. Like she's not complaining, she's, she's (child with POMS) like Okay so I'm just glad I found out what's going on with her and we can move on. I'm like, that's the way you have gotta approach the situation also. You can't sit there and think about the what if's. You know, you can't do that. So finally, once we got him on board, it went good from there. We was good as a family.

Judy shared the most compelling story regarding her husband and the impact that it had on him and their relationship as a couple and as a family. She began the conversation with the following:

> Between me and you, I was devastated, but it was very strange. Usually I'm the one that falls apart on things, and I had to be the strong one, because of my husband falling to pieces. I feel, and yes, I'm a very religious person, and if I feel that God made me the strong one in this, because my husband couldn't handle it, and my son, he knew that he had to hold it together. I wonder sometimes, did he

> hold it more together the first couple of years because he knew his dad had trouble with it?

During the interviews, the caregivers revealed that most of the younger siblings did not share their feelings because they did not understand the disease process due to their age. Sarah and Judy shared what they perceived as the feelings of two of the more mature younger siblings of the children with POMS.

Judy shared her younger son's reaction to his sibling's diagnosis of MS. She stated:

> This young man has always been well above his years. I'd say he has an old soul. He was very upset that his brother had been diagnosed with a disease that has no cure at the moment. He didn't know what to expect. He felt compassion for this brother…

The caregivers shared what they perceived as feelings of the older siblings of the children with POMS as well. Sue talked about the sibling of her child with POMS, who was six years older than the child with POMS. She stated, "My son, he worried about her, but he tried not to let it show and he still does today." Shannon, who recalled her oldest daughter's feelings of her younger sibling being diagnosed with POMS, "Her older sister, she said she felt helpless because she's that big sister, she wanted to protect her little sister and she can't."

Of all the caregivers, Lisa's family presented a unique perspective about the reaction of the siblings. Lisa stated that she felt that the two older siblings had the typical reaction of worry and concern for their sister but then their concern turned inward. Lisa added the following, "they had concerns about the disease being hereditary and their risk of having MS".

According to the Resiliency Model of Stress, Adjustment and Adaptation, not only does stress and strain include prior unresolved family strain and the family's reaction to the diagnosis, but it also includes changes in the family life cycle (McCubbin & McCubbin, 1983; McCubbin, McCubbin, Thompson & Thompson, 1995). The caregivers discussed the changes in roles, as well as the immediate, gradual, and unexpected changes that occurred within the family. They also discussed their perceptions of the how the changes affected the siblings of the children with POMS. This portion of the interview will be discussed in the subsequent sections.

Changes after the Diagnosis

The caregivers were asked how their role and the role of other family members changed as well as the amount of time that was spent caring for the child with POMS. Some of the caregivers reported that the roles within the family remained the same. One of the caregivers stated that the entire family took on a more active role and became more attentive to the child with POMS. Sarah stated the following in her interview:

> After the diagnosis, we all had to be more alert and that changed it because, you know, you were used to just going on. You really ain't paying attention. But after this, we had to really pay attention to (child with POMS). We had to pay attention to her surroundings, how she does things. Like, coordination, like cleaning up. Cause it's different. Like, when she's feeling well, one thing's one way. And then we have to just really pay attention to her motor skills and stuff. Because, as you know, it's hard with kids because they can hide it. Because, for one, being honest about it. So we went through the in denial part at first with it. So we had to really

watch and pay attention because she wasn't gonna say anything. So we had to be more alert.

Other caregivers talked about their roles changing to protector and advocate. When Judy was asked about how her role changed, she stated, "…I got more protective of him than I already was. Because he was already a special child. He, he had ADD and, um, and he struggled in school." She continued in her interview and discussed how she became an advocate for her child with POMS in school to prevent the child from failing but she also encouraged the child to work harder. She mentioned how her husband, who had previously been hard on their son, "took a step back to decrease the pressure so that he could figure out about his next steps in getting ready for college". Daisy talked about her role as an advocate for her child in the school setting as well.

Two of the caregivers, Sue and Morgan, discussed the various roles that their husbands took on after the diagnosis. Sue discussed how her husband became more sheltering of their daughter after her diagnosis. Morgan discussed how her husband, who is the stepfather of the child with POMS, became more involved in appointments and travel to clinical visits. Morgan also added that her spouse began spending more time with the child with POMS and in the decision making regarding his stepdaughter's care. One of the caregivers, the paternal aunt of the child with POMS, talked about how her role changed from the "fun aunt" to the "disciplinarian" with more of a paternal role since the decision was made for her nephew to come live in her household.

Two of the parents, Judy and Daisy, mention the word *caregiver* in their interviews when they talked about changes in their roles. When asked about the change in her role, Judy described:

> Me being mom, I was already a mom, a caregiver, but I had to step it up a notch, because once the diagnosis hit, then once we found a medication for him, we had to be trained on that, we had to administer that. That was a deal breaker, because he couldn't miss a dose. You had to make sure that it was done. It would be stressful….

Tina implied that she took on the role of a "caregiver" in her interview and stated that her "husband become more of a provider". Tina provided details of leaving her role as a full-time employee and her husband having to do overtime to maintain their income level. Lillie, the mother of child previously diagnosed with POMS, who now has progressive disease, spoke about her transformation from caregiver to overseer to prevent caregiver burnout:

> At the beginning, she was still doing a lot still on her own. But it started getting, I mean I've always been her mom, so I've always made sure she was good on everything. But you know as a teenager you don't really rely on your mother to do as much. You know, from combing your hair, to even brushing your teeth. You know, I kind of have to help her to do all that now… even the small things…she can't do on her own anymore. I really had to get some assistance for her, because it was really becoming too overwhelming for me.

Lillie also informed me in her interview that she employed an assistant for four days of the week and that her and her husband became overseers of her child with POMS care and provided respite care to her assistants. Lillie also reported that she worked most of the time since her husband has been unemployed; the caregiver reported that she awakens

each morning prior to work to perform personal hygiene activities and returns home in time before the sitter leaves.

One caregiver, Heather, reported that she has found a new role in mentoring other parents of children with POMS and other individuals with MS about the knowledge that she has attained to help others deal with their experiences.

When the caregivers were asked to elaborate on the immediate changes that took place because of the POMS diagnosis, nearly half of them stated that they could not identify any immediate changes that took place within their family. The other half of the caregivers discussed changes that included: 1) locating appropriate treatment and care, 2) recognizing and becoming more cognitively aware of the symptoms and their management, 3) accepting the diagnosis and 4) becoming more understanding of the child with the diagnosis and how they choose to deal with it.

When Judy was asked about the immediate changes that took place after the diagnosis, she stated:

> I was trying to grasp, to make sure that he (child with POMS) was seen by who could help us, and let us know how, what treatment was available, how to go about getting him treatment, and making sure he was able to live a successful life.

Lillie discussed her initial priority was getting her child "good doctor care". Karen added in her interview, "I think we became very aware and had to learn how to research medical stuff and how to deal with insurance stuff so that we could get his medication."

Several other caregivers discussed their need to recognize and become more aware of their child's symptoms and management. Nicole discussed how she felt the need to have someone constantly with her child with POMS to ensure her safety and how

she had a group of individuals, mainly family members she called if the need arouse to stay with the child. Daisy discussed in her interview the immediate need for the child with POMS to recognize "when she's feeling different, when things are not right, rather than just ignore it…" while Heather discussed how she had to personally "get herself together and not be panicking more and being stressed." She added the following insight:

> I had to be more alert, you know just to really pay attention to a lot of stuff. Make sure I don't miss nothing. Be aware of different signs of relapses or different things taking place that was happening to her that she probably wasn't realizing it was happening.

Several of the caregivers talked about their empathy toward the child diagnosed with POMS. When asked the question about immediate changes, Shannon revealed in her interview, "Immediate acceptance, accepting something that she … I guess accepting something new, accepting this new chapter…accepting this new thing (POMS) in our life that we gotta deal with …". Sadie discussed the recognition of her child's regression from independence and freedom to staying at home and conducting herself with hesitancy and eventually making better decisions about her health and her time. Sadie stated the following, "Me and my husband had to allow their child with POMS to grow and discover herself and what MS meant in her life".

While more than half of the caregivers reported that their families did not experience unexpected changes due to the diagnosis, the other half attributed their unexpected changes to school accommodations, unexpected medical expenses, the loss of independence and the recognition and management of symptoms and treatments. Most of the caregivers who reported unexpected changes discussed the recognition and

management of symptoms and treatments. Nicole stated in her interview, "I had to make sure that she is taking those shots. Regular. Make sure she has all her medications with her if she goes to her friend's house, I have to make sure she has everything with her at all times." Shannon elaborated on her experience with the unexpected changes:

> I guess, for me, knowing what MS is and knowing kinda, talking to I guess you guys at Children's and some of the different symptoms that she has been dealing with and we didn't know. Putting the symptoms and what she was dealing with…together with what we were seeing.

Donna shared her experience in the interview and discussed her unexpected changes regarding "a sense of what's to come" concerning doctor's appointments, treatments with medications, infusions and check-ups and all other necessary appointments. One of the other caregivers, Karen, revealed her family's unexpected expenses due to medical bills and her role to gain access to services and coverage that had not been utilized prior to her son's diagnosis.

The most compelling unexpected change that was shared by Lillie, the mother of child who was diagnosed with POMS at the age of 14. At the time of the interview, her child was 21 years of age, but Lillie perceived how, at the age of 18 only four years after her diagnosis, things began to spiral for her family:

> Her senior year in high school was when she really started getting the worst with her illness. She went from one relapse after then next. We was just having them back to back. Then we were coming to Birmingham every month at one time. We were coming from December up until they hospitalized her in March at UAB. And from her being able to walk on her own, to having a walker, because there

> was no way she could just walk without an assistant anymore. You know, so, it was very, very, very emotional. I think that when she got hospitalized at the end of the year, in that same year, they just said she just had to have a wheelchair. And I think that was the worst, because she never wanted to be in this chair. So, I think that's been the hardest part right now.

Some of the changes after the diagnosis were immediate, some were unexpected and yet some were gradual. Many of the caregivers did not perceive any gradual changes that took place within their families. For those caregivers that reported gradual changes, those changes were similarly identified as either immediate or unexpected changes and included the recognition of symptoms, the management of the disease, and the acquisition of school accommodations. Sadie and Karen discussed their personal needs as caregivers to "make things normal again" and to "become comfortable with the diagnosis". Sadie shared the following in her interview:

> The longer she's had it, I'm a little bit more laid back than I was early on, because I was always afraid if she overdid it, it would trigger something; didn't want her to be too far gone myself, you know, if something happened. Basically, learning to let her live her life despite my fears.

Changes that affect the sibling were brought up as well. Most caregivers perceived that the changes had no identifiable effect on the siblings, although some caregivers experienced feeling of guilt and neglect for their other children. A few of the caregivers witnessed what they perceived as "sibling animosity" toward the child with POMS. Judy recalled the last five years:

> I will say this, but now that I look back, I feel that maybe I shafted the younger

> brother, because our focus was upon the brother with the MS. At some point in our life we probably favored and catered to him and there may be some resentment somewhere from the younger brother. It was not meant and done intentionally, but as I look back, there may have been a little bit of that.

Joan admitted that she worried about the younger sibling of the child with POMS. She also shared her perceptions of the potential relief the sibling of the child with POMS would feel after the child went off to college and the center of attention no longer on the child with POMS. Sadie who had a younger child as well, shared her concerns:

> … and then you're worried; Am I spending enough time with the other child who is healthy? Is he going to start feeling like, you spend all your time with the (child with POMS) ...you're always running around with (child with POMS).

Two of the caregivers, Daisy and Katie, shared their personal encounters with siblings who openly expressed their feelings about their sibling with MS. Katie stated the following:

> Yeah, yeah, my daughter was really upset because I didn't get to spend Mother's Day with her. And, she was like, Well, uh, you're spending it with (child with POMS). You care more about her than you do me.

The child with POMS, who had been diagnosed a little less than one year, was recently hospitalized during a holiday and the sibling was not understanding.

When the caregivers were asked how much time they spent providing MS specific care, most of the them admitted that it was hard to quantify at the initial diagnosis because they had to take into consideration not only physical time but emotional time as well. Shannon, whose child was diagnosed nearly nine years ago at the time of the

interview, stated in her interview, “I guess when she first got diagnosed, for me, I was trying to wrap my mind around it 24 hours per day seven days a week.” She added, “It was 24 hours. But we constantly talked about it, worried about it.”. Nichole and Daisy who both have children that had been diagnosed less than one year at the time of the interview shared the same sentiments regarding time they spend caring providing MS direct children to their children with POMS. Denise, whose nephew was diagnosed less than a year, also agreed with the notion of constant care. She elaborated that “it was the medication, keeping track of his appointments with various specialists, arranging school and home bound services as well as his three flare-ups since the diagnosis and his ongoing emotional issues” that are so time consuming. The other caregivers admitted in their interview that it was less than a couple of hours per week unless there is a flare-up. Most caregivers admitted that their children learned over the course of the disease how to minimize the involvement of their caregivers.

In summary, the theme of “stress and strain” examined the demands of the families of children with POMS perceived by caregivers as they shared their experiences in the interviews with the researcher. The caregivers’ perceptions and ideas were expressed by direct quotes concerning any stressors that the families experienced prior to the diagnosis and during the initial phase of the diagnosis. In addition, the caregivers’ perceptions on the changes that took place within the family were discussed as well.

Adjusting to the Diagnosis

The caregivers in the study were asked to share their perceptions of: 1) the family’s view on raising a child with POMS 2) challenging issues related to the diagnosis; 3) the effects of the diagnosis on family relationships; and 4) the family’s

feelings about the future of the child with POMS. They were also asked to share descriptions of the children's expressed feelings about having MS. This information would reflect the caregivers' perceived appraisal of the degree to which caring for a child POMS has affected their family life based on the demands of the diagnosis and the caregivers and family's perceived capabilities. The capabilities of the caregivers and family are guided by the values, beliefs, and goals as well as priorities and expectations that are shared in their functioning (McCubbin & McCubbin, 1983; McCubbin et al., 1995).

The analysis of this portion of the interview resulted in the emergence of the theme "adjusting to the diagnosis" and consisted of five sub-themes, which were based on answers to the research questions. These sub-themes included the following: a) *family views on raising a child with POMS, b) future: optimism versus realism c) shared feelings about having POMS, d) family challenges, and e) family relationships after the diagnosis.* These sub-themes will be discussed in the following sections of this chapter.

Family Views on Raising a Child with POMS

The caregivers shared their views and their perceptions of the family's views on raising a child with POMS. Only half of those caregivers who were married had an open conversation with their spouse or other family members about this subject matter. The others formulated their own opinions about raising a child with POMS. Dollie, who was the mother of a child diagnosed over eight years ago, explained that her daughter's strength helped her to not feel "so helpless as a parent" and helped her as a mother to accept the diagnosis and all it entailed.

Sarah, the mother of a 16-year-old child that had been diagnosed for five years, revealed in her interview that she and her spouse had regular and open conversations about raising a child with POMS and talked about how the diagnosis can take an emotional toll on a family. She stated in her interview:

> It can be tiresome, it can be very overwhelming. But learning how to deal with certain things and not over act, you know when we first found out, we overacted over everything. But now, it's getting better over the years with how we deal with things. So, we talk about it all the time.

Two of the caregivers who have had some challenges dealing with their children diagnosed with POMS discussed their feelings of being overwhelmed. Both caregivers had unique experiences that drove their feelings about the subject matter. Denise, the maternal aunt to the child with POMS, talked about her decision to become her nephew's primary caregiver to assist her sister who had a major chronic illness as well. She talked about the struggle between her and her sibling regarding decision-making on her nephew's care. She also shared her perceptions of her family's views and opinions on how she dealt with her nephew regarding his extreme anger and behavior issues, which are direct effects of the disease manifestations of MS. She stated, "um, I think, we're much more lenient on him, because we want him to have that normal childhood, but he's not, 'cause he can't go to school, and so, I think he gets away with a lot more." She added that she felt the overcompensation causes issues of jealousy and nepotism among the siblings and others in the family as well.

Lillie, whose child was diagnosed at 14 years of age but is now 21 years old, shared her emotional experience of how she viewed raising a child with POMS. Her

child's disease has become progressively worse over the last three years and she expressed her struggles over this:

> I wish it would go away. That's not possible… It's really hard for me for real. I feel like I'm running from stuff, you know. I don't want to face a lot of stuff that I actually see. I don't really want to talk about it, a lot, because most of the time when I talk about it, it makes me very emotional…. What mother want to really see their child go through so much, you know? And you watch them go from one thing to the next and there's nothing you can do about it. You can't take it away; I wish it was me a lot of days. You know she is so young. Why does she have to go through this?

Future: Optimism versus Realism

The caregivers were asked to share their personal feelings and their families' feeling on the future of the children with POMS. A few of the caregivers stated that their families did not dwell on the future but lived day-to-day. The other caregivers shared their own optimistic view of the child with POMS future while others maintained a realistic view. It was observed that the caregivers' feelings of optimism were based on how the child progressed in their disease or the child's endurance and determination to achieve or do a task. On the other hand, realism was observed to be based on the knowledge acquired about the natural progression of the disease.

Sarah, the mother a child diagnosed five years prior to the interview, stated the following:

> Oh, her future is bright. She wants to be a…She loves culinary; she loves to bake with wheat. She wants to be an MS doctor. It's big. It's no limits. At first, we

> were scared like, what in the world? Will she ever get to move out? And now, were like helping her to do those things because we say how she looked at things.

Heather and Katie talked about their children's future being bright because of accomplishments their children have made despite having a diagnosis of POMS. Heather stated:

> Uh, I still feel her future is bright. She graduating next week so she's accomplished a lot from age 15. God knows what she is up to at 22, so I still say that's a lot of progress with research ahead of her and she still doing great. She able to do everything that she's been doing since she was 15, so I think it's, it's pretty good.

Despite the stabilization of disease in most children with POMS, some of the caregivers took on the realistic view about the future of their children. Maria stated in her interview,

> In my opinion MS as it is today is a progressive disease that could be, and I am going to put it straight out, that could lead to disability. I mean, that's a fact. It could lead to disability…the information that I remember seeing is that most MS patients within 10 years of diagnosis, they be on a cane. Within 10 to 15 years, they may be on a walker and then possibly a wheelchair. Even with all the medications out there, that is still how whether it's right or wrong, that is still how I see it.

Dollie, the mother of a child diagnosed with POMS at 14 years old is now 22 years and had two small children of her own, expressed her concern for her child as well as her grandchildren. She stated in her interview the following:

> Well you know she's got her babies now and I think about wondering if she's gonna stay able to take care of those babies. You know someday she gonna end up in a wheelchair or you know blind or any of the number of things that can happen to her. Just you know hoping and praying that doesn't happen so that she will be able to take care of the babies and herself and you know have a good life…I think about that a lot.

Donna, the mother of the youngest child, also expressed concern about her child's future, "I think we (her family) all probably worry a little about the damage...that's being done or you know, that he won't go back to his baseline." Other caregivers shared similar concerns about the unpredictability of MS regarding flare-ups, permanent symptoms, and disease progression. They also expressed their children's concerns about their future regarding their careers and family life.

Shared Feelings about Having POMS

The caregivers were asked if the child with POMS expressed any personal feelings about having the diagnosis of POMS. More than half of caregivers admitted that they had not had any open conversations with their child with POMS. Three of the caregivers, Joan, Tina and Heather, discussed their children's desire to "be normal" and live their lives like normal teenagers. At the time of the interview, two of those children had been living with a diagnosis of POMS for two years or less and one has been living with it for more than 5 years. Dollie, whose child had been diagnosed for 8 years at the time of the interview, restated her daughter's opinion, "This disease is not gonna control me." Shannon shared a similar response when asked about her child.

Nicole and Katie shared that their children expressed concerns about the realities of the disease over time and their personal embarrassment regarding symptoms they experienced with flare-ups. Nicole, whose daughter had been diagnosed for only a year at the time of the interview, discussed how her child with POMS shared her feelings of embarrassment. Nicole recalled the following:

> Now, when we first found out about it, she didn't understand what it was, so she didn't really have no feelings about it. The only thing that bothered her is that, you know, she was numb, and she felt embarrassed. Like, she couldn't go to ball games without trying to walk. She felt embarrassed. But now, as she's come along ... And she's still a little embarrassed 'cause a lot of her friends doesn't know that she has MS and stuff, but she's coming along pretty good.

Katie shared her perceptions of her child's experiences with POMS:

> I mean, she was getting to where she, like her left …it was her left at first. It was her left leg and her left foot. She could not feel it. And like she would, she couldn't …I mean, she would have to hold on to stuff to walk. And she, um, well, she was embarrassed to go to school, because she didn't want to walk down the hallways on a walker. She thought people were going to make fun of her. So, she went on home bound for a while.

Katie's daughter had been diagnosed less than a year at the time of the interview when she experienced these symptoms. Sadie, a mom of a child diagnosed with POMS for two years at the time of the interview, stated the child with POMS did not talk much about her diagnosis to her or her spouse but the child took on a different approach. She stated the following in her interview:

> She doesn't normally talk about herself, and she started a blog about her illness a little over a year ago, and it's posted, I think, six articles about it, sharing what she's going through with people and about how she feels like her illness has given her a closer relationship with God than she would've had without it.

Denise, the maternal aunt of child diagnosed with POMS less than one year at the time of the interview, revealed her observation of her nephew's feelings. In her interview, she talked about the symptoms he experienced and her perceptions that he felt "socially isolated". She believed that the flare-ups made her nephew feel hopeless but he did not share his feelings about it. She stated "Although, right now I think it's…He's in a bad spot, and I think he sees it as a, um, as a terminal diagnosis, although MS is not terminal."

Family Challenges

The caregivers were also asked to describe the biggest challenges that they had to overcome as a family since their children were diagnosed with POMS. More than half of the caregivers perceived that their families' biggest concern was the fact that their family had no control over the disease because of its unpredictable nature and uncertainty regarding the disease response to medication. When the other caregivers were asked their responses included the following: managing all the information about the disease, college plans, school issues, medical expense, insurance issues, administering shots, having multiple diseases, difficulty with communicating with younger POMS patients, physical and occupational therapy, and pregnancy with POMS. The most common challenge was the families' acceptance of the diagnosis. Dollie discussed her family's biggest challenge:

> Probably accepting the fact that she (child with POMS) has a disease that we don't' have any control over. That you know, we can't know…there's not been a treatment yet that you know kept her from relapsing, you know for very long. And you know just knowing that her, you know her MS is… It's never been clam, basically…we can't fix that. I can't fix that. And that was big, you know, even to this day that's still big, big deal to me.

Judy provided another perspective about the challenges faced by families of children with POMS. She stated, "I guess the biggest challenge was how to adapt, to how to make this part of our life, our daily life, to be able to live with it, and to expect, sometimes, the unexpected." Nicole discussed the "constant worry" as a parent regarding the medication working and her child with POMS' general health. She was also concerned about potential hospitalizations and the strain it may impose on her family financially. Judy stated the following:

> That her shots is not gonna work. They're not going to help her. That, you know, her left side …She was so numb, that if she get like that again, she's going to have to be in the hospital, and that going to put a strain on finances. I be trying to work and not take off.

Joan added that her biggest challenge was her son's compliance with his medication and the fear of how his adherence with his DMT would affect the future. She stated the following in her interview:

> The biggest challenge is, one of them, um, dealing with the fact that you have a six-teen year old child with a disease that you don't know what is going to do with him…And you don't want to be pounding. You have MS, and you've got to

> take your medicine. Um, and that's been a challenge, is making him understand he needs to take care of himself even though he feels well.

Other caregivers discussed their child's permanent and temporary symptoms and how those symptoms affected their family's life. Carol recalled her child with POMS most recent flare-up. The child was in a wheelchair due to her MS symptoms and the caregiver remembered he worry over child being incapacitated for the rest of her life. Although this was a temporary symptom, Carol was fixed on the possibility of it happening again and becoming a permanent symptom if the medication did not control her child's MS from progressing. Denise shared her story regarding her nephew's cognitive and personality changes when asked about challenges the family faced over the past year since his diagnosis. Denise stated the following:

> Personality changes in (child with POMS). Um, well because I'm with him a lot, I can read him easier. And a lot of times, because of the brain being affected, and the cognitive issues he's had, the extended family doesn't always see it. And then when he does have a flare-up or if he's having an angry moment, just trying to help them realize that it may be where the lesion is in his brain and it's the MS.

Family Relationships after the Diagnosis

The caregivers were asked to describe how the diagnosis affected their family's relationships with other people within the home as well as those relationships outside of the home. Concerning relationships within the home, most of the caregivers revealed that they believed the diagnosis brought them closer as a family and strengthened the bond between them. Only a few stated that the diagnosis had no perceived effect on their relationships within the house. Dollie shared the following in her interview,

> I think that brought us all closer when she got diagnosed because we knew we had to come together to make sure that she got where she needed to be and you know really watch her and really do what you know she needed us to do.

Shannon shared that she felt as though her family began to value each other more and that they pray together more because it had more value and meaning to their lives. Joan also revealed a similar concept in her interview, "We were very close. I mean, I would say a very close family…I think the diagnosis has probably made us closer and more appreciative of our family."

One caregiver, however, revealed how she believed the diagnosis changed the dynamics of her relationship with her child who was diagnosed with POMS. Maria admitted that prior to the diagnosis her relationship with her child was typical of a son being raised in the home by a single female parent. After the diagnosis, she observed changes, but she could not determine whether those changes were due to the diagnosis or him needing an active father figure in his life as he transitioned from a teenager to a young adult. Because of her domineering style of parenting, she felt that the relationship between her and her son changed. Maria shared the following about her and her child's relationship in her interview:

> Before his diagnosis? Pretty good. I mean, for a teenager, it was yeah. We still do have a pretty good relationship. Things are changing with us. I don't know if (child with POMS) shared with you, but this summer I've let him stay with his dad for a couple of weeks because he needs to be with a male figure and we were king of butting heads sometimes…I think it was good for him. Because he needs to talk with a male figure. He needed to talk with somebody who understands.

When asked about the changes she perceived, she stated the following, “It’s different. It’s different because when he doesn’t talk I think something is wrong or if he’s sleeping a lot, I’m thinking is something wrong. You know?” Maria admitted she felt her experiences with her child with POMS has taught her to be more of an active listener. She added:

> I need to listen to him. I mean, when he’s talking or not talking or not talking because I look at him a lot. He notices that. I do need to listen because you can hear but you need to listen.

Judy, another caregiver talked about her relationship with her husband. She discussed how she believed it became strained after her son was diagnosed with POMS. She stated the following:

> I was angry with him (my husband). Because he was supposed to be the strong one, the rock, and how dare he…He was so angry, he was so mad with God, that God allowed this, and to his son. Obviously, I was mad at my husband, because he was mad at God…For us to have been so fun-loving and happy-go-lucky, all that, and then this to be a bombshell…There were days that there would not be talking in the house.

She added that because her husband remained angry and in denial regarding his son’s condition and its effects. It took nearly two years before their relationship returned to normal.

Another caregiver stated that she believed her relationship with her child with POMS was strengthened. However, the relationship with the sibling of the child with POMS became strained because of the attention that the child with POMS received due to

her illness. Katie tearfully talked about how the younger sibling of the child with POMS appeared to feel left out and stated, “she’s kind of jealous of it”. Katie’s child had multiple flare-ups that occurred consecutively and required a lot of time and attention from the caregiver, which caused less attention to be on the sibling. Katie shared that the younger sibling expressed her feelings openly with her maternal grandmother, which brought on a lot of tension and anxiety for the caregiver as a parent.

Several caregivers stated that they believed that their relationships outside of the home with their extended family, church family, and community grew stronger. Those relationships became sources of support for caregivers as well as the child with POMS. Nicole talked about her family’s closeness and togetherness after the POMS diagnosis. She stated the following in her interview:

> Everybody’s, like, participating more. We talk more. They try to show up as much as they can. Like I said, if I’m at work, someone will take her to her appointment for me. If I don’t understand something, I can call my aunt or her…My husband’s sister, and they can explain to me…So, outside the home, they are a great help.

Shannon, Lisa, and Joan also expressed how they felt the diagnosis strengthened their relationships outside of the home including their extended family, church family, and friends and built a certain amount of comfort despite the diagnosis.

Sarah described the perceived change in her immediate family’s relationship with her extended family and friends. She attributed it to the fact that her immediate family’s actions are often misunderstood. She revealed the following:

> Sometimes it comes across as…We don't have a whole lot of friends, if that makes sense. And we do have some relationships with family but it's different because…its different because they don't seem to understand why we do the way we do. And we come across as hypochondriacs….so it's kind of difficult. They're supportive. They don't understand. And they're supportive in their own way so we have to change the way we looked at our friendships. We had to change the way we looked at family relationships and friend relationships.

Two of the caregivers perceived that their child being diagnosed with POMS led their families to develop more compassion for individuals with disabilities. Karen shared her family's experience:

> I think we have become more understanding of people with disabilities and in particular people with maybe some medical things going on that you can't see. Because we've learned you can't see it and there's a lot going on with people. I think we have become more giving of our time. Understanding and giving.

Heather also observed a similar family experience, "… we ran into some other people with MS and that helps us to talk to them. And you know, they scared like we was when we first found out so that's a good thing." She added it was a "good change" to be able to pass on her family's knowledge to others about how MS can be manageable.

In summary, the theme "adjusting to the diagnosis" resulted from the analysis of the caregiver interviews on shared perceptions of the families of children with POMS regarding their views on raising a child with POMS and the challenges they faced as a family. Additionally, the analysis revealed the effects of POMs on familial relationships and expressed feelings about the diagnosis of MS on the family's current life and on the

future. These factors were examined to determine the family's overall appraisal of the diagnosis.

Communication

The caregivers in the study were asked to describe how their family communicated with each other prior to the diagnosis of POMS. They were also asked to describe how communication within their family changed during an unexpected medical event related to the child with POMS (i.e., an MS exacerbation or flare-up). Family problem solving communication can be a predictor of how a family adapts to a chronic illness diagnosis. The theme "communication" resulted in the analysis of the interviews, with three sub-themes, which included: a) *daily mode of communication and b) changes in communication and c) communication during a relapse.*

Daily Mode of Communication

Some of the caregivers agreed in their interviews that they believed their family communicated well with the people within the household. Nicole, Heather, Katie, and Sadie shared that they talked to their families throughout the day and had discussions either in person or by phone or text, depending on where they were and what they were doing. Nicole stated,

> All the time. Every day, all day. If I'm not at work…She's in my face…If I'm at work, she's calling me on the phone. If she's at her friend's house, calling me on the phone. So, yeah, Me and her, we talk a lot.

In contrast, Nicole added that she and her spouse, whom she was not living with at the time of the diagnosis, had broken communication with each other and she attributed that

to separation in living arrangements and not the diagnosis of MS. On the other hand, Sarah stated in her interview that her family joked with each other to make the conversation and communication more interesting. When asked to describe how her family communicated with each other daily, she said, "Through joking. We try to keep each other lifted. We talk serious but we also keep the laughter. We try to keep the laughter going."

One of the caregivers, Karen, shared that she talked to both her children all day because they are home schooled and they rarely go anywhere without her. She stated that she and her husband communicated via text message or phone call. Sue and Judy admitted that their family communicated some throughout the day as well; however, their family had discussions during dinner or in the living room. Sue provided this insight on her family's communication in her interview:

> As I said earlier, we try to meet, round-table discussion, at least three nights a week for supper. I am in education, so I'm off this summer, so a lot of days it's just me here at home. The youngest one has a young lady in his life, and so they going and doing. Now my (child with POMS) has him a young lady, so it's like four adults living in the house. We're all going in different directions most days. At least three nights a week, we're all together. We're all working and just doing life.

While most caregivers reported that they felt that there was open communication among their family, not all caregivers reported feeling the same way about their family. Lillie shared the following: "That's something we need to work on. Because I think I've shut down a lot. I don't do a lot of communicating, you know, sometimes. I really

don't." Denise, the maternal aunt to the child with POMS, elaborated in her interview that she believed that sometimes communication is difficult because her nephew's anger and cognitive issues prevented him from communicating with her. Denise also felt that her sister, who is the mother of the child with POMS, struggled with communication because of her recall issues related to her chronic illness that causes short-term and long-term memory loss.

Changes in Communication

Most of the caregivers stated that they believed there were no changes in communication after the diagnosis; however, some talked about perceived changes in family communication that were observed after the diagnosis. These caregivers talked about silent communication, communication by observation, increased communication with others outside of the home, more open communication, more effective communication and yet some talked about closed communication. The following paragraphs will discuss some of changes in communication mentioned above.

Dollie revealed in her interview that she felt that she and her child with POMS had open communication prior the diagnosis but that communication changed after the diagnosis. She explained, 'I would just ask her, but she didn't…prior to the MS she was I mean if something was going on with her… I'd just ask her and she would tell me." Dollie felt her child became less engaged in the communication between them and became more secluded and isolated after the diagnosis. Dollie revealed the following,

> After the diagnosis that changed a little bit. She wouldn't…you know she would have times where she would just lay in the bed and I could just tell by looking at her face something was wrong and she would never tell me. She kind of clammed

> up and wouldn't talk. You know and I mean never did find out you know anything from her like how she was feeling about it. But you know when she was younger she just clammed up. Things kind of changed you know. I think because she didn't want me to get upset…Prior to that we didn't …She would just tell me you know if anything was going on. She would talk to me about it.

Sarah revealed a comparable situation she observed with her family regarding communication. She perceived that her family used silent communication to process their situation. She stated the following:

> The communication sometimes, it'll get silent. Cause I think everybody's trying to process, okay something's different. But then once…Then everybody slowly comes around and we'll slowly talk about what's happening. We try to get through the day of it first. Then the next day, then we say, oh this was different. "

She admitted that she believed communication changed from day-to-day based on the situation at the time. She added "We joke and laugh and talk and then like, silent. Okay, we gotta deal with something else…And then we talk about it, yes." Nicole and Tina perceived that their family's communication changed after the diagnosis to include more openness about how the child with POM felt.

One of the caregivers shared how her family's communication changed since the diagnosis. Lilly shared her experience regarding communication with her child with POMS, her husband and her overall communication with others. Lilly's child with MS was diagnosed eight years prior to the interview. She stated that she felt family communication was strained and admitted the child with POMS didn't like to communicate with her as her parent. When asked why she felt her child with POMS does

not like to talk to her, she stated the following: “(Child with POMS) says she don’t like to talk to me. She talks to her dad ‘cause I get too emotional and it make her (child with POMS) emotional.” Lilly added the following when asked about her communication with her husband, “Sometimes. Sometimes we don’t either.” She admitted the following about her communication overall and how it has been affected by the diagnosis:

> I really don't know. I... I have... I don't know because sometimes I feel like I'm running from stuff, you know. I don't want to face a lot of stuff that I actually see. I don't really talk about it [MS], a lot, because most of the time when I talk about it; It makes me very emotional, so I kinda like, just... I think I just make myself kind numb to the situation. Because sometimes (child with POMS) be like, Well, Mom, how does this make you feel? And I'm like, I have my days, you know," but my days are most times when I'm by myself, I'm at work, I'm in my office and I get to thinking, and I think about where she was, and how things once was, and the things she desires to do that she's not able to do. I guess sometimes that gets to me, and she don't know that it does, because I really don't share that with people. And I don't really show it either, you know.

Communication during a Relapse

Almost all the caregivers admitted that communication during a relapse is very task oriented and driven by what the child is experiencing and how as a family they are going to deal with the situation. Communication appeared to be more frequent whether it is by phone, text, or in person with the common goal of finding a solution to the situation they may be facing. Judy described how she handled communication during a relapse:

> I feel like I go into overload mom, of…And my husband too, he goes into overload dad. It's like, what can we do? It's going to be this, this or this. What's going to be the best step to take first? We'll ask him (child with POMS), just say he's on a steroid each day, okay, did her feel better today? Can we move that leg better today than you did yesterday? Is it better on scale from one to 10? Yes, we amp it up.

Sadie shared her experience, "We get on the phone and we start talking, and working things out to make it happen."

Yet some communicated less than others did during a relapse. One of the caregivers, Karen, admitted during her interview that she "retreats within herself "and does less communicating. She also added that although she is doing less communicating, she remained "task oriented" and "focused on her child's health."

In summary, the theme "communication" resulted from the analysis of the caregiver interviews on shared perceptions of the families of children with POMS regarding communication prior to the diagnosis of POMS within the family and changes in communication after the diagnosis. Additionally, communication during the time of a relapse, which may be a time of crisis for some families, was discussed a well. This theme provided knowledge about how families communicate as a means of problem solving and adaptation to resolved issues that arise.

Coping with the Diagnosis

The caregivers were asked to describe what strategies the family used to cope with the ongoing challenges associated with raising a child with POMS. Family coping

are those strategies, patterns and behaviors that strengthen the family and help them to maintain the stability and well-being of its members (McCubbin & McCubbin, 1983; McCubbin, McCubbin et al., 1995). A family managing day-to-day may be different from managing during a crisis to deal with a diagnosis of POMS because a crisis or time of uncertainty may bring about a change in the family's coping ability. From the interviews, the theme "coping with the diagnosis" emerged with the two sub-themes: a) *day-to-day life and b) changes during a relapse.* These sub-themes are discussed in the following sections.

Day-to-day Life

Most of the caregivers in the study reported that they managed day-to-day life by normalizing their life and establishing a routine. Most of these caregivers had children diagnosed two or more years with POMS. Tina, Lisa, and Karen admitted that they took life "day by day". Heather stated her family focused on "staying positive" and "moving forward" despite the diagnosis. Sadie and Sarah confessed that their families live by a schedule when things are running smoothly and it is a non-eventful day. Sadie stated the following in her interview:

> When things are running smooth, just stick to the schedule. I'm very Type A and I like everything planned out. If we're going to stick to a schedule, you know, of who's going to be where, or who's taking (sibling of child with POMS) to school, who's picking him up; (Child with POMS) got that little part time job so I know exactly when she's coming and going. Unless she's having a bad day, everything kind of runs like a well-oiled machine, unless something happens.

When Nicole was asked how her family managed day-to-day life, she stated, “I say we just manage through our prayer. We pray.” Joan and Morgan discussed how they proceed throughout their day with care and caution. When Joan was asked the question about how her family managed their day-to-day life, she stated the following personal convictions about her child’s condition:

> It’s a little different, ‘cause I feel like I have to be more mindful, um, paying attention to how he feels, and you know, if it’s cold, is he dressed well, more, because of sensitivity, if he’s hot, if he’s hydrated? You know, I’m constantly trying to, um, make sure that he’s taking care of himself.

Joan also admitted that her day-to-day life is filled with worry and concern about how her son will independently take care of himself when he goes to college. Morgan added the following perceptions, “I would say probably just because we’re more careful and we’ll try to pay attention if she doesn’t feel well and all that.” Since the diagnosis, both families appeared have learned to become more attentive, careful, and observant of their child with POMS.

Donna admitted in her interview that although her family went about their day as usual, she worried about her child with POMS going to school and his possible inability to communicate if he was experienced a symptom. Donna’s child with POMS was 7 years old at the time of the interview.

Although most of the caregivers’ experiences with day-to-day life were not always challenging, a few caregivers faced challenges almost daily. When Denise was asked to describe the day-to-day routine with her nephew and family, she described the following, “It’s really hard. It’s a -emotional rollercoaster.” She elaborated on the

numerous flare-ups that her nephew experienced in the recent past and the emotional imbalance of their lives because of the unexpected symptoms. Lillie also shared a similar experience. She acknowledged that her day-to-day life considering her child's progressive disease. She stated the following when asked about her strategies for day-to-day life:

> If I have any? I ... um. I don't even know, most of the time, it's always her. I think that's... It's crazy but I'm always trying to make sure she's good even if she doesn't see that. Um. Yeah, even when she doesn't see that.

Changes during a Relapse

More than half of the caregivers reported perceived changes within their family or themselves during a relapse. Tina, Katie, Carol and Karen all reported that their children with POMS required more help from them as their primary caregiver; however, everything else and everyone else operated as normal. Lillie admitted that because of the severity of her child's most recent flare-up, she became more emotional than she usually does. She stated,

> …I get kinda, I think the last time she got really sick, it really scared me really bad. We had to call the paramedics for her, that freaked me out. I couldn't even. I just about went numb or something, because I just sat there. I couldn't deal with the situation.

Denise stated in her interview that her nephew had experienced three flare-ups since his diagnosis and that each flare-up was different in symptomology and severity with different treatment regimens. Denise added that she and her sister shared the

responsibility of seeking outpatient medical treatment and therapy for her nephew and staying with him during times that he was hospitalized.

Sadie and Sarah discussed that although their families lived by a daily schedule, the child with POMS became the center of attention when a relapse was experienced. Sarah provided an example of the family changes that occurred when her child with POMS is in a relapse:

> Everybody kind of pitches in. Her sibling runs around helping. If I'm dealing with (child with POMS) more, he dad picks up supper, or help more with the housework. It's like everybody kind of knows that there's going to be a lot going on, she needs a lot of help, so you know who's going to take her to the doctor, and working our schedules around making sure she gets everywhere she needs to be; school or wherever it is.

Nicole's child was recently diagnosed with POMS, and Nicole admitted that she worried more during a relapse. In her interview, Nicole stated that she believed her family worried more because there was so much they did not understand. She stated,

> I'm still learning. So, I try to take it one day at a time, but I really don't know how to feel because so many people tell me so many things and then I google things, Is she this? Is she that? So, it's really hard.

Judy also admitted that despite her child having POMS for five years, she was worried whether her child would recover from the relapse or have residual symptoms.

Shannon, whose child was 13 at diagnosis but is now 22, described how things have changed during a relapse now versus when she was younger. She admitted when her child was initially diagnosed at age 13:

> It was hard. For the first relapse it was hard, cause like I said we always seen the little stuff. But to see the actual effects of MS, what it can actually do. It was hard…It was an adjustment. It was hard for me. I cried. I didn't want her to see me cry, so I hate to see her go through that, but she was a trooper.

Shannon's child with POMS remained at home at age 22 and Shannon admitted the following:

> When she has a flare up, now that her being an adult, she stays at home and most of the time, my mom will come check on her. We'll constantly call and FaceTime, whatcha doing? How you doing? But she's at the point where she's driving, she can go to the doctor herself. If it's really bad, me and my husband will go with her, but other than that she goes by herself. And we try to let her be independent on her own. She's 22, so it's hard, but I gotta let go.

Donna shared her struggle with a child with POMS during a relapse because of his age and his difficulty with expressing his feelings. He was seven years old at the time of the interview and was diagnosed at age four. She stated the following when asked about her specific struggles during a relapse:

> …Being understanding to some of the issues he was dealing with, but something I still struggle with, even now…is not letting it be an excuse for other things, like if he's feeling bad, … I know he feels bad, but he can't act ugly or adjusting to that. Not letting it be an excuse for behavior.

In summary, the theme "coping with the diagnosis" resulted from the analysis of the caregiver interviews on shared perceptions of the families of children with POMS regarding the family's management of day-to-day life. This theme also revealed how the

function of the family changes during a relapse. These factors were examined to determine what strategies, patterns and behaviors are used to assist families during a time of crisis.

Sources of Strength

Family resources can serve as a source of strength for the family as a unit and can influence the capabilities of each individual family member (McCubbin & McCubbin, 1983; McCubbin, et al., 1995). These resources aid families in managing stressors and strains that a chronic illness diagnosis may cause. Additional adequate resources such as acquired knowledge and support help to restore balance in the lives of families who may have otherwise limited resources (McCubbin & McCubbin, 1983; McCubbin, et al., 1995).

The caregivers in this study were asked to describe their current level of knowledge about MS and how their knowledge and understanding of the disease has changed over time. Caregivers were also asked to share their perceptions of their children and family's knowledge and how this knowledge changed over time. Asking these questions would address the family's personal strength. In addition to those questions, the caregivers were asked to describe any resources (i.e. medical, community, personal, technology) that assisted their family in raising a child with POMS. Although there were no specific questions regarding faith and religion asked in the caregiver interview, it was clearly perceived by the caregivers as an integral part of the support and resource for the caregivers and their families. The analysis of this portion of the interview resulted in the theme "sources of strength" with 4 sub-themes: a) p*ast and*

present levels of understanding, b) resources to improve knowledge, c) role of faith and religion and d) social support. Each sub-theme is discussed in the following section.

Past and Present Levels of Understanding

All the caregivers in the study admitted that their initial level of understanding was very limited because either they did not know anything about the disease, or they only knew what they had seen in the media (i.e. television, internet, etc.). Their worries included concerns about permanent disability, loss of independence, early death, being bedridden, wheelchair bound, and limited options for the future. Dollie whose child had been diagnosed nearly eight years at the time of the interview was asked if her understanding changed over time. She stated the following:

> "…Yeah it's changed since her diagnosis 'cause you know there's a lot of things I didn't know about MS that I know now. So you know that her MS is not going to kill her. You know I just understand it more. I know what it is and what it does and how it affects you know people who have it."

Although Katie's child had been diagnosed with MS in a year at the time of the interview, she stated the following:

> Oh, yeah. Yeah, it's, it's a different understanding. Um, because at first it was just like, uh, you know, I thought she, well she's gonna be, uh, bedridden, and she's gonna die at a young age, or whatever. But now, it's like, "No, she can live, you know, a long happy life with it," I mean with, with the right medication.

Sarah, whose child had been diagnosed with POMS for nine years, admitted in her interview:

> It was just like a new normal, you know. We got new information and we just felt like it was a different direction that God allowed to come our way. So we looked at it a little different.

When asked why her understanding changed, Shannon shared that her acquired knowledge about MS brought her to a new awareness of several things such as her daughter's struggle with taking injections. She also became more aware of individuals with chronic illness and their emotions. Shannon shared the following experience:

> I do. Cause I mean, when she first diagnosed the shot, the Rebif shot, it was awful. I just couldn't understand why she wouldn't take it and get it over with. And we battled. I mean every three days. Three days a week, we battled. I cried, she cried. I had to understand, that you know, it's hard for her. So, I had to start taking shots for rheumatoid, and I got it. You know, it's hard to stick yourself with a needle. But now, you know, I listen to her. It was hard, cause like I said for me, if you're not going through something, it's easy for you to say what you really want to do. So, I couldn't understand why she was being ... why you acting like this? You gotta take it, just take it. But, now you know I just listen to her and try to put myself in her shoes. Like I said, she's a trooper, so. It's much easier now, than it was then. So, I try to keep an open mind, and we pray together a lot.

In addition, Shannon admitted that she came into an awareness that her daughter's struggle with MS was mild in comparison to others and that she must learn to be grateful for her daughter's experience. She stated the following:

> I think me meeting people. Talking to people that have MS. I have met a lot of people that have MS. Listened to others and how they cope with it. Some people

> that's going through stuff, but you know more severe than others and you're looking at your child and I feel selfish for feeling bad about it …for her. I know people are going through things worse off than her.

Sadie and Denise, who both have children diagnosed less than two years prior to the interview, admitted that perceptions of their knowledge waxes and wanes from one day to the other. Sadie admitted, "I'm going to be honest. The more I think I know, the more I realize I don't know, you know? It's like I'm constantly learning new things." Denise stated:

> I feel like, um, it depends on the day. Sometimes I feel like I know everything. I try to explain it to everybody, and then, when it's just me sitting at home. I know nothing and I need to research everything.

Denise also added that the focus of her knowledge changed based on the situation her family and her child with POMS were in at the time.

When the caregivers were asked about their perceptions of their child with POMS' level of understanding about MS, they reported comparable stories. Most of the caregivers reported that they believed their children with POMS initially did not have any knowledge of what the diagnosis meant. However, they believed that over time the children with POMS began to develop an awareness and understanding through their reading, support groups, internet research and talking with their healthcare providers. Most of the caregivers reported a perceived improved level of understanding in the child with POMS if they had been diagnosed two or more years.

Dollie and Lisa reported that their children with POMS developed what they viewed as an awareness of their knowledge of their disease process as time has passed. Dollie reported the following in her interview:

> She's just grasping that. She is learning her body; she knows her body. What she can and what she cannot do." So, she's at the crossroad now to her future. She's thinking more now about her future. About her being a wife and children and what effect MS is gonna have on her being a mother and having kids. That's her biggest thing now.

Lisa reported a similar experience when she revealed her perceptions of her child with POMS:

> I do think she understands it better. I think she understands that she has to let us know if something starts bothering her. She has to be more in tuned with her body. I do think she is more in tuned with her body.

Lisa and Dollie had children that had been diagnosed with POMS at 6 years and 8 years respectively. Shannon reported that although her child with POMS "learned her body" similarly as Lisa and Dollie's children, she stated that her child perceived herself at a "crossroads" about her future and the decisions she will make regarding her life. She stated the following:

> I think she understands it, but she's at the point now, she's 22, and she's thinking more about her future, like a husband and that kind of stuff. She's at that crossroad now, where she's confused. She talks about having children now. So that part, she understands what she has.

Sarah reported that she believed her child with POMS understood that each person with MS is affected differently. She admitted teaching her child that there is no comparison between individuals with MS and their experiences. She stated the following in her interview, “What may affect one person may not affect her the same way. So I think that kind of helps her get through any challenges that she has.”

Donna, the caregiver that had the youngest child (7 years old) diagnosed with POMS, stated that she did not believe her son had a good understanding of what an MS diagnosis means due to his developmental level but that he connected his disease with the lack of use of his legs. She stated the following:

> He associates it with his legs. Um, that was his ... It wasn't his first flare up, but it was the major one and it was the one he got the diagnosis um, during that time. So he thinks that MS makes your feet mess up, 'cause um, we slowly ... I'll bring it up and we try to talk about it. Um, and I you know, I guess we try to keep it on his level. And he- he kind of tells you, I guess as he grows, I kind of think- ... he can understand a little bit more, but right now he just knows that um ... He does know that there's lesions on his brain and spine. And that those spots are what sometimes make his legs tired or- ... whatnot, and that um, his medicine is to prevent him from getting more of those.

Resources to Improve Knowledge

Nearly one-third of the caregivers used the internet as a resource to improve their knowledge regarding MS, although most caregivers admitted the information is limited regarding POMS. All the caregivers in the study identified the staff at UAB’s Center for Pediatric Onset Demyelinating Disease (CPODD) and/or their primary care providers as

a resource utilized to improve knowledge about multiple sclerosis. CPODD was noted as providing direct teaching and advice to the caregivers and their families as well as opportunities to collaborate, network and improve their educational knowledge with other families at the annual retreat event.

Nearly half of the caregivers identified the National MS Society as a resource utilized by families to improve their knowledge as well. Specifically, Maria, Shannon and Lillie identified pamphlets, magazines, and resources such as durable medical equipment as items physically obtained from the National MS Society. In addition, Sadie verbalized that she and her family had the opportunity to participate in support groups and the MS walk, which are National MS Society sponsored activities designed to raise awareness of MS and the community that is involved to provide services for persons diagnosed with MS and their families. Sadie added in her interview that although she saw the support group as beneficial, her child with POMS did not want to continue participating because most of the individuals attending this activity were at least 25 years older than she was. Sadie also identified the local vocational rehabilitation as a resource for high school accommodations and job placement after high school.

Sue identified the Pediatric MS support group as an on-line Facebook® resource that she and some other parents used and Sarah stated that some of the mothers have connected through email after building a personal network to connect to other families. Sadie and Maria acknowledged the pharmaceutical manufacturing companies that provide injection training and on-going teaching are a source of patient education and support as well a source of other company sponsored client and family activities.

Nicole and Denise felt like their families did not have any resources to connect to

in the community. They did not live near a pediatric MS center nor did their child have a pediatric neurologist experienced in providing care for the child with POMS. Denise also added that she also previously used the internet and the National MS Society in a limited capacity as a resource. Denise stated the following in her interview:

> (laughs) it feels like there are no resources. Just fighting for everything, because trying to get the medication approved, it takes months and months and months, because everything is approved for adult MS patients, and there's nothing approved for pediatric MS. Um, in Tampa area, we have no pediatric neurologist doctors who deal with MS, so, it's a fight. We were trying to get him an EEG last week and it took us six weeks to get that scheduled, because the hospital didn't have a pediatric neurologist who could read it.

Role of Faith and Religion

Nearly half of the caregivers discussed the role of faith and religion as a personal resource to help them deal with their child's diagnosis. Sarah identified how she perceived her family managed an unexpected crisis through the power of prayer to give her family direction. She stated the following,

> We [are] overwhelmed. But then we immediately turn to prayer. We turn to prayer and then we try to sort out, okay now how are we gonna do this? How are we gonna…What's next? And then we tend to have a plan in place. Like if somebody would need the car, if it's something, we tend to have it kind of together.

Likewise, Nicole and Tina described that they believed their families utilizes the power of prayer to manage the effects of the disease as well. In addition, Tina talked about her

mother's faith that "God has the power to heal her child with POMS". Judy talked about the "inner strength that God gave her at the time when her child with POMS was diagnosed because her husband was angry because of the diagnosis".

Sadie recounted a conversation her husband had with her to comfort and ease her fears associated with the diagnosis:

> Then of course, my husband was like, (caregiver), you know, she's in God's hands. He's got a bigger plan than we can see; we're really just going to have to give it to Him, even if it's not a day-by-day thing, even if it's a minute-by-minute thing. Just turning it to God and let Him take that worry, and knowing that He's going to put her in the right doctor's hands, and that whatever's meant to happen will happen.

Sadie also recalled that she believed that every time their family went through a crisis, her faith became stronger. She revealed:

> …I feel like my faith has become stronger too, and I've learned more about...versus I guess like, getting emotional and keeping it. I've learned how to literally lay things at God's feet, like he wants me to versus keeping it an trying to figure it all the answers on my own.

Sadie added that her grandmother who lives eight hours away, had intercessory prayer at her church for her child with POMS and that a prayer chain was started to show support. Daisy admitted in her interview that sometimes she and her child with POMS and the sibling read scriptures together to offer a sense of what she perceived as peace to the family. She added that if things became too difficult at home, the child with POMS would go to her pastor at church for guidance, reassurance, and prayer.

Social Support

Less than half of the caregivers identified social support that helped them with the physical demands and challenges that the diagnosis may pose on the family of a child with POMS. Sue, Heather, and Sadie identified the grandparents of the children with POMS as the main source of social support when it comes to the sibling of the child with POMS. Sue informed me of her parents' role in providing care for the sibling of the child with POMS so that she can travel 100 miles one way to the neurologist for treatment. Heather stated the following about her mother, "I, once again, thank God for my mom. She was there. She didn't have a problem with keeping him, with getting him, none of that." Sadie added that she had the support of both her parents and her husband's parents. She revealed that both sets of grandparents often made special trips when the child with POMS was having a bad day and brought special dishes when the child was not feeling well.

Donna, who lived with her parents at the time of the interview, agreed that they were a major source of support for her and her child with POMS. She also identified friends as well as her sibling outside of the home who were a source of support for her and her child as well. Katie acknowledged that her siblings and her husband's siblings helped with her other children when they needed to attend to MS appointments miles away. She also identified her church family, friends, and her network of people with MS as means of social support for her and her family. Karen admitted that her family lived nearly two hundred miles away; however, "her church is a big resource for us."

Joan and Lillie were the two caregivers that identified siblings of the child as the main source of support for the child with POMS. Joan stated the following in her

interview:

> I would say his big brother immediately stepped up, um, and was very supportive. He took on, um, being, uh, there for child with POMS, and, um, and trying to show him support and love. And, um, and we-we just tried to show him that he's going to be okay, and this is not going to change who he is, and we just all kind of just were, just rallied around him in trying to show him that we loved him, and MS is not going to change him, and, and that, uh, he still has a full life. So, we all just kind of, just stepped up.

Joan admitted that although the sibling of the child with POMS did not live at home and was away at college, he made extra trips home and frequently communicated with his sibling when he was initially diagnosed.

Joan added in her interview that she had the support of "some really close friends" but some were not so supportive. She revealed the following:

> And the ones you didn't hear from, you're like, 'Okay, they're not going to be your friend. They don't really care.' So it's weird that it's really sounds cliché, but it's true, it like the people step for you; Really you know who they, who loves you and care for you.

Similarly, Lillie discussed the support of the older sibling of the child with POMS who did not live in the home. The sibling and his spouse moved from their home two hours away back to their hometown for six months so that he could help with his sibling. Lillie also revealed an emotional response regarding the lack of support of "true friends" for her and her daughter:

> And that's why I try to instill in her, yeah we be emotional, yeah we cry, I cry too. I hurt for her. I hurt because you know I have friends, you know, people you thought was friends, but when you really get down, you really find out who your friends really are. She don't have people come by and say "Well, (child with POMS), I'm gonna take you for a ride." If she goes anywhere, it's with me. You know what I'm saying? It's not where she just actually goes and hangs out with friends, that don't happen. She hangs out with me. You know most of the time we go to the movies, she with me. You know, it's just. It's very rare if she goes out with a friend, my niece might come take her somewhere, or do something with her. But that's you know rare because they have their own lives. So, it's me and my baby most of the time. It just be me and her.

Judy was the only caregiver who identified two sources of support from her community. One of the sources was a local pharmacy in her area that they had not previously patronized in her family's hometown. Her child with POMS needed a powdered steroid during a relapse but her regular pharmacy could not provide the medication. She shared her experience:

> I'd like to share this one story though. He had a relapse, and you all wanted him to take, it was a powdered steroid. One pharmacy could not get that, so we have another pharmacy in our town that was able to provide us with that service. To have you know, we do not shop ... I shop there for candles, go figure, but they still ask about him. From that one time. To me that's a community outreach, because they could have denied, since we were not patronages to them, they could have denied that, and they chose to help us, and to still ask about him.

Judy went on to discuss, the football team and her child with POMS' other friends as a source of support for her son during that same flare-up. She recalled how her son's teammates and friends would surround him and cheer for him while he drank his oral steroids that he thought was so horrible in taste. The child with POMS was the quarterback of his football team and his classmates wanted to show their support to him. Mom concluded, "I would say that's a community."

In summary, the theme "sources of strength" resulted from the analysis of the caregiver interviews on shared perceptions of the families of children with POMS regarding their views on resources that serves also a mechanism of coping and adapting to the diagnosis of POMS in families. In this analysis, past and present levels of understanding in the caregiver, families and child regarding MS was examined as well. In addition, the role of religion, social support and the resources that improved families' knowledge about MS were discussed as well. These factors were examined to determine the family's overall capabilities that empower them to adapt to the demands of the disease process.

Achieving Balance

According to McCubbin & McCubbin (1993), successful family adaptation occurs when the family can achieve a balance between the needs of the individual with a medical condition, the needs of the family as unit and the needs of each family members. The caregivers in the study were asked to share strategies that developed as a family to manage the demands of the illness and the needs of the family. The theme "achieving balance" resulted from the analysis of the interviews.

All except two of the caregivers identified strategies that their family utilized to achieve balance and to make a positive move toward family adaptation regarding the diagnosis of POMS. Families who seemed to be moving toward adaptation utilized a variety of strategies to achieve balance and adaptation. The findings will be discussed in the following sections.

When asked about their family's strategies for balancing, Dollie, Judie and Lisa discussed their perceptions that their families' biggest strategy was to live for the moment and not dwell on the diagnosis. Dollie stated:

> I don't have anything planned out when something happened you know. We would just call the doctor and take off to the doctor or you know do whatever we had to do. I didn't really have a strategy laid out.

Judie eluded to a similar strategy in her interview; however, she admitted that in the beginning after the diagnosis was made, she believed her family "prioritized the need to try to fix and to heal, to get rid of this disease". She stated that after a year, the family developed a new perspective in which they "take it day-by-day". Julie further described that she thought their main goal was for all family members to make it through the day and start a new day tomorrow. Lisa shared a similar approach, "I try to live in the moment and look a little bit ahead, but not too far ahead. I really don't think…We don't know…MS is such a different disease for everybody."

Sue, Sarah and Sadie agreed that their main strategy was to balance the needs of the siblings and family and not focus on the child with MS. Sue discussed in her interview how she did not want her other children to feel left out so she planned special events in which she spent time with the siblings exclusively. Sadie agreed that it was

important to make time for each child, while managing time better and reevaluating situations based on their level of importance. Sarah concurred that she did not allow the child with POMS to interfere with the desires of the other family members. She also discussed how she believed her family recognized the manipulative behavior of the child with POMS when it came to identifying MS symptoms.

On the other hand, Nicole and Lillie discussed that they put the need of the child with POMS before everything else including themselves. When asked if she thought their family was balanced, Lillie stated the following:

> I don't think so at all. Like I said, I just drop everything. It's not even about my needs…What I need to do, when it comes down to her, it's her. Um, sometimes maybe it's stress...I might have to go and take a ride, or whatever. But I still, my main focus is her.

Joan admitted that she believed her family's focus was on the child with POMS; however, she wanted to ensure the child with POMS understood that there were other family members in the home to consider. Heather revealed that she focuses on herself and her personal health so that she could maintain the health of her children including the child with POMS. Of note, Heather is a single parent.

Tina stated that she believed her family's main strategy to achieving balance was family togetherness. When asked what their strategy was, Tina stated the following:

> Doing things together. So, if (child with POMS) is down and out for some reason or super tired or something, we really will get together and watch, have a family movie night. The strategy's just doing stuff together.

Karen shared a similar strategy in which her family spends a lot of time together and engages in open communication as well as family prayer.

While Tina and Karen discussed family togetherness, Katie discussed her perceptions of her family's top strategy of "family help" in which she depends on her family outside of the home to aid her and her family when necessary. Carol and Shannon perceived that their family's top strategy is the belief that their families need to function as a team and work together to achieve a common goal no matter what it is. Similarly, Morgan talked about how she believed her family discussed any situation they faced and came up with a solution as a strategy for their family.

Maria, who lived in the home alone with her child with POMS at the time of the interview, described how she allowed her child to spend time with his cousins who were his age as a strategy for achieving balance. She decided to do this to prevent him from feeling isolated and to establish a sense of normalcy in his life despite the disease and its manifestations. She admitted that this allowed her time to get a "mental break" and clear her mind as well.

When asked about her family's strategies, Donna stated the following:

> "All of us just getting back to a normal life once you get the diagnosis and learn everything that you can to try and deal with it and then…. incorporating that into your normal routine. Um I think it kinda goes back to not allowing the MS diagnosis to rule everything."

Of all the caregivers in the study, Denise and Daisy both indirectly stated that they did not have any strategies that they believed help their families to achieve balance. When asked the question regarding her family's strategy of achieving balance, unlike the

rest of the caregivers, Daisy discussed a list of demands that prevented her from developing a strategy to maintain balance. Some of the demands had been mentioned previously and some had not. She discussed the demands of balancing her time between the child with POMS and her other child. Daisy stated the following:

> I'm not going to lie. That's difficult for me because I'm learning now because my son just told us how he feels last night. So, I have to hear him. I mean, I don't just hear. I have to really listen to what he's saying. You know, he's hurting. He's hurting that, you know, he has a sister that he's dealing with. I know that she's sick. I know I see the changes. I see. She talks to me. I know this. So, I'm trying to get that right now. I'm in the process of that right now. So that's a little bit. Now I'm trying to find some time for him where it's just he and I. That's difficult because it's just the three of us. So, where we go, Angel has to go with us. I don't like leaving her. That part's difficult, but we did decide that we wanted ... I did tell him. I said, "Let me take you to the movies," and we just wanted to do something fun for us. So, we'll just make it whatever you wanted to do day. So, we're doing that this weekend just to relieve some stress for him because sometimes ... Not sometimes. My mind, and my whole focus, and my whole attention is so centered on (child with MS). Looking out for her, and no dating, and you know, on top of this I'm trying to sell my house. All of this, and I'm being pulled in many different directions. He gets left. So, he makes sure that his voice be known. That, "Hey, I'm important too. Please take my needs into consideration as well, you know." So, I heard him yesterday. With tears in his eyes, he voiced that to us, so we got it. At least we can do something for him as well, so yeah.

Daisy voiced being pulled in so many directions regarding her personal health, selling her home, school issues for the child with POMS and her other child the physical and mental abuse of her and her children by her spouse weighed heavily on her emotional state.

In summary, the theme "achieving balance" resulted from the analysis of the caregiver interviews on shared perceptions of the families of children with POMS regarding strategies utilized by families to balance the demand of the disease with their life as a family. The finding within this theme allowed the researcher to examine how they reached some sense of normalcy despite the diagnosis.

The Overall Experience of the Family

The caregivers in the study were asked to provide a synopsis of their experiences on raising a child with POMS by imagining that they were writing a book. The caregivers were asked to identify a title and main idea of the book. The caregivers were also asked to identify any advice they would give to other families of children with POMS who have children recently diagnosed and to give advice to healthcare providers who care for children with POMS and their families. The analysis of the information revealed the final and seventh theme, "the overall experience of the family" which includes the following sub-themes: *(a) book title with the main message (b) advice for families and (c) advice for healthcare providers.* The sub-themes will be discussed in the following section.

Book Title with the Main Message

Near the completion of the interviews, caregivers were asked to sum up their experiences by identifying the title of a book and the main message about family

experiences with raising a child with POMS. Most of the book titles conveyed messages of hope and positivity for families of children with POMS like the “The New Normal”, “Making the Good of the Bad Experiences “and “Enduring”. And yet, other titles were references to goals and expectations for families of children with POMS such as “Overcoming Life with MS” and The Challenges that You Face with a Child with MS”. The summary of the messages in the book were directly related to the title and how life as a family should be viewed and lived when faced with a diagnosis of POMS in a child living in that household.

Advice for Families

The caregivers were asked to share advice with other families of children with POMS. There was a wide variety of responses. Some shared their thoughts and opinions about the parental role and yet others gave suggestions about the need for changes in the family’s role. The advice that caregivers shared is discussed in the following paragraphs.

Shannon and Dollie suggested that parents become knowledgeable about their child’s disease but suggested the avoidance of the use of the internet. Dollie stated that the best sources were from professionals and Shannon agreed that parents should not always believe the internet as a factual source. Shannon added that the “fundamentals of MS” should be learned from health professionals. In addition, Shannon advised to “live day by day, pray about it each day”, and not to operate under pessimism when dealing with an issue. She also added that parents of children with MS should encourage their children “to love and respect their body”. Sarah agreed with Shannon and Dollie that parents should listen to those people that are skillful and knowledgeable as much as they

can although it may be overwhelming. Sarah added that each family should identify what works well for them and make decisions based on personal goals.

Maria also suggested that parents of children with MS research information about MS but suggested that parents should not get overwhelmed with the use of the internet although she did not suggest any other sources for parents to seek this information. She also added MS should be viewed as a "side thought" and not something that dominates the family's life. Donna agreed that families should learn as much as they can but she also advised that families should seek help and support from others when needed.

Tina, Dollie and Lisa advised that families maintain open communication at all times. From her experiences, Tina perceived that children are more open and acceptable to situations than their parents and parents are more "close-minded". She suggested, "Parents need to take a deep breath and let go and let the child with POMS do their own thing". Dollie advised to keep the communication between the family and child open so that the family is aware of what is going on with the child all the time. Similarly, Lisa advised maintaining open communication while staying informed and taking a minute to take everything in without panicking.

Daisy offered different advice to families to consider when listening to their child with POMS regarding their symptoms. She offered the following statement in her interview:

> My advice to a parent that I hear a lot when I was talking with the neurologist is that parents don't listen to the children. The children say that there's a concern, the parents just brush it off. My advice is to listen to the concern of every symptom, and investigate every symptom, and also watch. Listen and watch.

Although, Sadie did not suggest advice on listening to the child with POMS, she recommended that parents and families of children with POMS seek advice from individuals who have gone through similar situations. Sadie also added advise to seek from others about the disease without feeling ashamed about their concerns or lack of knowledge. She shared the following in her interview:

> Just having somebody to talk to, because parents with healthy kids don't understand. They don't get it. They don't understand why you drop everything and run, they don't understand when your child, to look at them physically on the outside, they seem fine and they're not. You know, they don't get it.

Karen and Katie offered two very different kinds of advice to families of children with POMS. Karen suggested that families spend time "grieving about the diagnosis and the loss of their normal child" in order to find the "new normal". Katie advised families to make every attempt to "accept the diagnosis" and the value of what it means to their family. She also suggested that parents be willing to provide support for all the children in the family, including those siblings of the child with POMS so that all the children receive the support and attention that they need. In contrast, Lillie insisted that parents of children with POMS learn to find time for themselves to prevent becoming "lost in the child" and lacking in the area of self-care. She shared the following:

> The main message I think I would want to give parents is, from what I see as a parent, is learning how to find time for yourself, because you really need it, because you really do. I think that's what I'm starting to do more so than I've ever done before within this last year. You know, coping and dealing, because

sometimes you dealing with it, and sometimes I think we forget about ourselves.

We put their needs so much greater and forget, and I've lost myself a lot, for me.

Lillie also added that families should stay positive no matter what they are facing and avoid “people that bring you negative energy”.

Advice for Healthcare Professionals

The caregivers were also asked to share advice that would give to healthcare providers who care for children with POMS and their families. The caregivers reflected on their positive experiences with their current healthcare providers and staff. The information below reflects their advice and responses in the interview.

Lillie advised that healthcare providers of children with POMS should have compassion for their patients and families and passion about what they do as professionals. Lillie reflected on her experience with her healthcare provider at CPODD. When asked in her interview, she stated the following characteristics she felt were necessary for that of a healthcare provider taking care of a child with POMS:

> A care provider to have love and compassion for what they do, and it shows through what they're doing. It’s so important when you're actually dealing with people with different illnesses. Everybody doesn't have that just because they're a health provider, they just don't. You know, when you put yourself out there where you actually like, plenty of times that I called you with my own personal issues, and you listened. It wasn't like you said so in so…., I don't want to talk to you today. You always made that time, you know what I'm saying, and it's so important.

Lillie also stated that health professionals should take time with their patients and families and not rush to provide care. Likewise, Joan and Tina added that healthcare professionals should take the time to get to know the family and child and help the child to understand how the disease effects a person. Joan added that it is important to talk to the children on their level about their disease process so that they can comprehend. Joan, Katie, and Donna also mentioned healthcare professionals should maintain open communication, be understanding with children and families and support their efforts as a family while maintaining honesty during their interactions in personal and professional settings.

Maria and Daisy provided additional advice for healthcare professionals. Maria advised that providers should ask their pediatric patients more questions about their feelings because oftentimes children are not forthcoming with the parents. Daisy agreed that healthcare providers should actively listen to the child and build rapport with the child and their family as well. Daisy added that healthcare professionals should allow children and teens the time to express their feelings so that they are comfortable with telling healthcare providers anything.

Karen and Carol addressed her advice for healthcare providers from a unique perspective. Karen stated the following:

> I think ... Advice for healthcare providers ... Understand the psychology behind it and what it does to a family as far as how they're gonna have to try to redefine what is normal for them and understand, I think, that it really is a grieving process and understand that the family is grieving, however long that's gonna be.

Karen's focus for healthcare professionals was to understand how MS affects the family and how it redefines "normal" for them. Carol's advice was for health care professionals to help children with POMS and their families to connect to others dealing with the disease through peer support for both children and parents.

Morgan and Sarah who both lived greater than 200 miles from a specialist and struggled to find local healthcare providers who were knowledgeable about MS, offered advice to inexperienced providers providing care to children with POMS and their families. Morgan stated that inexperienced providers should do more research about MS and become more educated about delivering care for children and families with POMS. Sarah suggested that inexperienced providers should be more open to building relationships with the clinical experts and be more willing to maintain open communication and seek opportunities to learn while receiving advice from the experts.

In summary, the theme "the overall experience of the family" resulted from the analysis of the caregiver interviews on shared perceptions of the families of children with POMS regarding families' summaries of their past and present experiencing with raising a child with POMS. This analysis also provided advice for other families and healthcare professionals who may seeking care for their newly diagnosed children in the future.

At the time of the interviews, some families appeared to be well adjusted to the diagnosis of POMs and yet some were still adjusting. Families, who display signs of maladjustment, may display signs of maladaptation and ineffective coping, whole those who are adjusting well appear to adapt and develop ineffective coping skills after the diagnosis of a chronic illness. Exemplars of a well-adjusted family and a maladaptive family will be shared in the upcoming sections. The discussion of a well-adjusted family

of a child with POMS will display how families learn to adapt after a diagnosis by utilizing newly established patterns of functioning, external support and other problem solving skills. On the other hand, the exemplar of the maladaptive family will offer insight into the diagnosis of POMS in which family demands, current family function, appraisal, and vulnerabilities could easily supersede adaptation if resources like newly established patterns of functioning, external support, coping, and problem- solving strategies are not put into place.

Exemplar of a Well-Adjusted and Adapted Family

Tina is the caregiver of a child diagnosed three years prior to the interview for this study. Tina discussed her family's life prior to her child's diagnosis that may contribute to a pile-up of demands. She discussed typical life changes such as increased work hours and job related demands and stressor like on-call hours, weekend work shifts, and difficulty sleeping. Tina also discussed her concerns regarding her oldest daughter starting college and the changes in family roles prior to the diagnosis. The oldest child had previously transported the child with POMS to and from school as well as other various activities and was no longer available.

Tina shared her family's response to the POM's diagnosis: "Um, when we first got the diagnosis, it was a relief and scary at the same time". Her family was relieved that "it wasn't brain cancer" but that they finally had a diagnosis for the symptoms child with POMS had been dealing with for the past two years. Tina also shared that she felt her family's emotional responses to the diagnosis were related to their fears of the unknown (i.e. manifestations, symptom management and treatment options). She also

stated, “I think the uncertainty of everything is how the emotions come flooding in…” Tina verbalized the shared parental responsibilities between her and her husband to balance their lives, even prior to her child’s POMS diagnosis. The family made a decision to change roles shortly after the child with POMS was diagnosed. Tina began to work less hours and became more active in the life the child with POMS, advocating at school of the child with POM and managing more of the care of the child with POMS. She also, with the help of her other family members, focused on modifying the family’s home environment to meet the physical constraints of her child with POMS. Tina’s spouse began working extra hours to offset the finances, but remained actively involved in the decisions and discussions regarding the child’s care. The sibling of the child with POMS was also involved in the discussion regarding her sibling’s diagnosis as well and often missed social activities to share in the family’s experiences of adjusting to the diagnosis. Although Tina shared her husband was very emotional for several months after the diagnosis, she also expressed that it did not interfere with their marriage or his responsibilities as a father. The family modified their lives as well to accommodate the child with POMS but made attempts to “normalize” the family after the diagnosis. Tina stated the following:

> If we do to the beach we’re gonna have to go to like later afternoon I think that was the way like how we kind of like changed our focus on how, as a family, like how we was gonna do things.

Tina discusses the overwhelming support she had from her parents and her spouses’ parents as well as other extended family members, but admits that she did not utilize any community resources other than her church family and the doctors and nurses

that care for her child. She explained that her family had strong religious beliefs and admitted that she felt that the power of prayer had a role in her family's response to the diagnosis. She also shared that the child with POMS had a very positive attitude, and she felt this attributed to the family having a more optimistic outlook on her child's future.

The characteristics of a well-adjusted, adapted family include: 1) their innate ability to be resilient 2) their adaptability to change, and 3) their desire to seek social support. Resiliency is a major premise in the Resiliency Model of Stress, Adjustment, and Adaptation and is integral in the initial discussion of a family well adapted in the face of chronic illness (Cardoso & Chronister, 2009).

Resiliency is the capacity to overcome situations and circumstances of adversity (McCubbin et al., 2002). In the face of chronic illness, a resilient family like Tina's family deals with the challenges of the illness and work together as a unit to manage the illness and meet the demands of each family member despite the pile-up of stressors. Coping is the process by which families engage in direct responses to demands and exhausted resources and come to the realization that changes need to take place to restore function and improve satisfaction (Cardoso & Chronister, 2009). Tina's family developed and practiced new coping strategies in order to achieve balance and fit at multiple levels including the individual to family and the family to community. Each member in Tina's family made an effort to deal with the illness and the changes that have taken place as a result. The family's adaptability to changes also measures coping.

According to the Resiliency Model, adaptability occurs with the formation of newly developed problem-solving skills and coping techniques that assist in making the crisis manageable (Cardoso & Chronister, 2009). The family will reframe personal and

family priorities in order to meet the demands of the child and the illness. Families like Tina's, who begin to adapt to the diagnosis, function with confidence and optimism and operate with a sense of spirituality and cohesiveness while sharing rituals and routines that promote close family relationships even during the time of crisis (Cardoso & Chronister, 2009). Techniques of adaptability may include role flexibility and maintaining day-to-day normalcy, humor and laughter and open family communication about concerns and frustrations. Tina's family practiced role flexibility in which they made adjustments with situations that arose regarding the illness. They also maintained open communication to provide clarity, an opportunity of expression for emotions and collective problem solving. These techniques along with the family's choice to seek social support created a sense of optimism and balance in the face of adversity.

Seeking social support will aid in the family's journey toward adaptation (McCubbin et al., 2002). Social support can be informal (family, friends and neighbors) or formal (healthcare professionals, politicians, employers and school officials). Social support can be viewed as a buffer and mediator between stress and psychological well-being (McCubbin et al., 2002). Support networks can be individual, familial and community based (McCubbin et al., 2002; Kelly & Ganong, 2011). Individual based support networks include a parent's education and income. Tina, the caregiver of a child with POMS possessed enough knowledge as an individual to advocate for her child at school. Family based social networks include the involvement of immediate or extended family in the care of the child. Extended or immediate family in Tina's family made a commitment to providing support. Tina identified the Annual CPODD family retreat as a community based social networks where she has the opportunity to share with other

families with similar experiences. Seeking support from other parents or advocacy groups that engage in the same efforts when faced with the challenges of a chronic illness can have positive benefits. Tina actively engages with her healthcare providers to gain a clear understanding about the disease. Fears of the unknown become minimal when families come to the reality that they have individuals who can alleviate their misnomers about the disease (McCubbin et al, 2002).

Exemplar of a Maladjusted Family

Daisy is the caregiver of a child who was diagnosed one year prior to the interview for this study. Daisy discussed her pile-up of demands as she began her interview. Daisy was asked to discuss her family's experience with receiving the diagnosis. She stated the following:

> Upon receiving the diagnosis (POMS), it was very, very difficult. The difficulty of knowing that what's difficult for me, and I'll try not to get teared-eyed, is that I have fibromyalgia and I have my own health concerns. I have chronic pain and so the difficulty for me is as a mother not being able to care for my child. I don't like to be vulnerable. I don't like to be weak. With the diagnosis that I have, sometimes it hurts to be touched. Sometimes my body aches and hurts really really bad, but if she needs me to assist her, my biggest concern is that I won't be able to provide the assistance that she needs because of my own disability. That's what frightens me. Another thing that frightens me is I don't want her to end up in a wheelchair.

Daisy's immediate concerns regarding her child with POMS was based on perceptions of her possible inability to care for her child. She then goes into a discussion regarding "discrimination based on disability" against her child with POMS in the school setting:

> The discrimination that I see with her is when she was able to ... When she went to school just prior to she's going into high school when she's 14, we were diagnosed, so it was all new. She was at home, online, doing her schoolwork at home online. She wanted to go to high school. She researched her school and we went and looked at the school and we weren't diagnosed at that time. She researched her school. She started having these symptoms. Yeah, the diagnosis we were told we just take this piece of paper in with the diagnosis and everything would just be hunky-dory. Everything would be perfect. Everything would be okay. Wave this paper and this is the answer to all our prayers, so I thought, or so the neurologists made me think. When we walked into her school and I showed them the diagnosis, they seemed somewhat sympathetic. They seemed like they were going to provide all the accommodations, but the moment I left the school, the accommodations left when I left.

Additionally, she discussed the difficulties at the school regarding accommodations despite an individualized educational plan (IEP) in place. She talked about the anger that she felt as a caregiver who was unable to convince the school that her child needed homework and test accommodations despite her daughter looking physically well.

Although this caregiver stated the she had the knowledge necessary to advocate for her child in a school setting to attain accommodations, she did not fully understand the need for school officials to be educated about multiple sclerosis and its effect on her,

the child with POMS, and other children. She had difficulty conveying that oftentimes symptoms are invisible in children and oftentimes affect the cognitive and emotional state of the child. Her appraisal of this situation was that the school discriminated against her child.

Because of the stress of school, the child with POMS became depressed and because of the depression, she began to inflict self-harm to release the negative emotions felt. Daisy recounts the following:

> Then she got so depressed she went into the bathroom, and she found a razor, and she cut herself pretty bad. We went to the primary care doctor and that's when she said that her disease is affecting the way she is able to function. The way she thinks. Her cognitive thinking. Now she's seeing a psychiatrist. So she's not seeing a therapist anymore. Now with the high anxiety that she's faced and that she's been under for so long, they've prescribed a medication for her.

It was observed that Daisy jumped from subject to subject throughout the interview. She had great difficulty focusing and often did not answer the research questions directly. When asked about stressors and strains of the family prior to the diagnosis, she shared the following:

> Prior to the diagnosis, we've been dealing with stress. I've come from a divorce now. was married to my husband for over 18 and a half years. The last two/three years of that was sheer hell for us. In 2008, my mother died and that sent all of our worlds crashing down. (Child with POMS) loved my mother, her grandmother so much. She was just like her mom.

Daisy shared an encounter with her ex-husband and the child prior to the divorce:

> So, he comes in at this time and says we're going fight. The kids are not knowing what he wants to do. He's pushing me around. He is high on something. So it became very problematic for us. It developed into a domestic violence. It developed into violence with me, violence with the kids. I kept going in and out. I was in severe depression. My mother was, you know, gone then and at that point I didn't really have a reason to live. So, some parts I remember, some parts I don't because I was too bad to remember and I don't know.

Daisy went through a difficult divorce after her husband's years of infidelity as well as his physical, verbal and mental abuse toward her and her children. Daisy also shared her feelings of loss and grief and the perceived feelings of grief of her children regarding the death of her mother after witnessing her mother's long battle with cancer.

Due to the divorce and the caregiver's recent disability, the family was forced to move due to financial strains. The children began to display outward signs of dysfunction. The child with POMS began to self-inflict wounds on herself while the other child displayed outward signs of anger and emotional outburst at home as well as in public.

According to the Resiliency Model, the family's vulnerability is determined by a pile-up of demands within the family unit such as debt, poor health of a member or multiple members, changes in work status, and changes in the family's life cycle McCubbin, McCubbin & Hamilton, 1993). Daisy's family is vulnerable based on the pile of demands on her family prior to the diagnosis of POMS. As a direct result of her prior experience, this caregiver was challenged with the initial diagnosis of POMS, which may be the direct result of her assessment of her and her families' prior experiences. She

feared her ability to be able to care for her child with POMS because of her own diagnosis as well as all the other factors mentioned in the preceding discussion. In addition, her child with POMS had experienced multiple flare-ups during the first year of the diagnosis and she had residual symptoms and well as cognitive effects related to POMS.

According to the Resiliency model, once families are introduced to a new stressor, they begin to use their family resistance resources, which are the family's ability and capability to address and manage a stressor and its demands while balance life to avoid a crisis (McCubbin, Thompson & McCubbin, 1988). These resources include things like social support, economic stability, shared spiritual beliefs, open communication, cohesiveness and flexibility. Daisy talked about her struggles with additional issues such as: 1) her child's primary healthcare provider who was not as knowledgeable about treating POMS; 2) the distance from the center and the lack of transportation to make regular visits; 3) the sibling's struggle with the attention given to child with POMS; 4) her child with POMS struggle to fit in with her peers due to her diagnosis; 5) the child with POMS displaying maladaptive behaviors such as cutting despite psychotherapy; and 6) the increased issues of ineffective communication within her family since the diagnosis of POMS.

If Daisy's family does not manage her child's disease and does not develop ways to problem solve and cope with ongoing stressors related to day-to-day living, the family will continue to maladjust and be at risk for crisis. Developing new patterns of functioning while eliminating those ineffective patterns of coping would be beneficial to the Daisy's family. In addition, Daisy's family must also be able to identify family

resources like community, family and social support that will allow change in her family to move into adaptation.

Summary

Twenty female family caregivers participated in this study to explore from their perspective how family factors influence adaptation in families of children with pediatric onset multiple sclerosis. The socio demographic data was analyzed. Seven themes emerged from the thematic analysis of the interviews. The themes were stress and strain; adjusting to the diagnosis; communication; coping with the diagnosis; sources of strength; achieving balance; and the overall experience of the family. The exemplars were written to illustrate well-adapted and maladaptive families. In the context of families dealing with a child diagnosed with a chronic illness. The next chapter will include a discussion of finding, strengths and limitations of the study, as well as implications for practice and recommendations for future research.

CHAPTER 5

DISCUSSION

The purpose of this qualitative descriptive study was to explore the caregiver perspectives of family factors that influence adaptation in families of children with pediatric onset multiple sclerosis (POMS) seen at a southeastern specialty center. Twenty family caregivers shared their thoughts and firsthand experiences of how their families lived from day-to-day and adapted their lives to care for their children with POMS. Family caregivers reported on family factors such as stress and strain, roles and responsibilities related to the diagnosis, views on raising a child with POMS, relationships, communication, coping and resources. Caregivers were given an opportunity to share not only their feelings, but also their perceptions of feelings of others in their family, including their spouses, children with POMS, and other children and adults living in the household. Families of children with POMS represent a rare population faced with a chronic illness that usually presents in adulthood. The findings from this study represent first of its kind research specific to the adaptation of families of children with POMS.

This chapter presents a discussion of the findings from this study as well as the study's conclusion, strengths and limitations, and implications for practice and future research.

Key Findings

Data analysis revealed many factors that influenced the adaptation of families of children with a diagnosis of POMS. Key findings emerged from the data and will be discussed in the upcoming paragraphs. The key findings are as follows:

1. For some families, the presence of a pile of stressors or atypical stressors prior to the diagnosis of POMS affected their perceptions of the demands of caring for a child with POMS.
2. Families' pre-conceived thoughts about POMS affected their initial reactions to the diagnosis of POMS.
3. Families of children with POMS who identified resources (i.e. community, spiritual and support) for coping better adjusted and adapted to the diagnosis, yet some struggled due to the perceived lack of resources.
4. Families of children with POMS with new-onset and unstable disease struggled with adjustment due to lack of knowledge about disease management.
5. The developmental needs of the child with POMS influenced the adjustment and adaptation of families.
6. Changes in family roles and responsibilities related to the diagnosis negatively affected some younger siblings of children with POMS.
7. The lack of communication among families of children with POMS related to issues with adjustment and adaptation.
8. The ongoing health status of the child with POMS directly affected balancing day-to-day family life.

Discussion of Key Findings

The stress on a family of a child diagnosed with a chronic illness like POMS can place demands on the family that may be new and challenging (Compas et al., 2012). The demands on these families perceived by the caregivers included but were not limited to changes in roles and responsibilities of various family members. Caregivers reported that some of them had to change their job status or obtain another job due to the number of flare-ups the child with POMS experienced. This was due to frequent doctor's visits or hospitalization in the initial period after the diagnosis when the disease was very active. Caregivers of children with POMS also reported that the adult family members often had loss of time from work during the initial period due to travel to the specialized center. Most of the families lived greater than 100 miles and traveled between one and three times per year to the center for routine visits and additionally for visits due to flare-ups or increased disease activity. Some caregivers reported that some of them along with their spouses filed for family medical leave (FMLA) with their employers to protect their jobs.

Findings from this study and similarly indicated in the Resiliency Model suggest that a recently accumulated pile of stressors or atypical stressors prior to the diagnosis of POMS could affect family perceptions of the demands of the diagnosis of POMS (McCubbin et al, 2002). Caregivers reported in their interviews that they were dealing with events like surviving a devastating hurricane, dealing with grief after multiple miscarriages, and being diagnosed with a major illness that could affect their ability to provide the care of a child with POMS. These events along with the diagnosis of POMS in a child in the household produced a great deal of anxiety for the families. In a recent review of literature, evidence suggested family demands of adults living with relapsing

remitting multiple sclerosis (RRMS) and supported that initial and ongoing demands on families could negatively impact family functioning (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016; Diareme et al., 2006; Paliokosta et al., 2009). However, recent literature on families of adults with RRMS does not support evidence that prior stressors affect a family's ability to function or cope as a unit. In one study young child caregivers of adults with MS acknowledged that their responsibilities were enormous and that they had limited professional assistance in dealing with their parent with MS which led to personal feelings of being "invisible and unacknowledged" (Bjorgvinsdottir & Halldorsdottir, 2014). Other studies found that needs of the healthy parent were being neglected and that children experienced anxiety and wory related to their parent's illness (Bostrom & Nilsagard, 2016; Diareme et al., 2006; Paliokosta et al., 2009).

According to the Resiliency Model, prior stressors can cause a pile-up of demands that may cause additional family dysfunction and impede adjustments to the diagnosis (McCubbin & McCubbin, 1983). Healthcare providers of families of children with POMS must consider the social and psychological needs of families after a diagnosis of POMS is made that may be related to prior stresses and their previous life experiences. These stressors may leave families of children with POMS vulnerable to additional stress, decreased family functioning, and in turn, leads to maladjustment (McCubbin et al, 2002).

In addition to the typical or atypical family stressors experienced prior to the diagnosis of POMS, most families of children with POMS are in shock and disbelief due to the initial diagnosis of POMS. The caregivers in the study reported that some of the

families of POMS were surprised at the unexpected news and while others were relieved to have a diagnosis after a year or two of unexplained symptoms. The caregivers also reported that these families immediately formulated ideas in their heads because internet information created a picture in their minds of how this disease affected adults in the past. Findings from this study suggested that the 'families pre-conceived thoughts about MS and its effects could influence their appraisal of the disease. Caregivers also reported that most families of children with POMS exhibited worry and dismay about the unknown effects on their children as they enter adulthood. Recent literature on families of adults with RRMS supported the fact that family appraisal along with shared beliefs, rules, values, and goals, shaped perceptions of the diagnosis of MS (Bostrom & Nilsagard, 2016; Fallahi-Khoshknab et al., 2014). These authors also suggested that this appraisal could create false perceptions due to inadequate information about the disease and its clinical manifestations. According to the Resiliency Model, the level of a family's ability to cope in response to a chronic illness is based on several factors, including the positive appraisal of a situation (Cardosto & Chronister, 2009). Prolonged adoption of negative family appraisal in families of children newly diagnosed with POMS due to misconceptions may lead to ineffective coping and maladaptation (McCubbin et al, 2002). Several studies regarding families of adults with MS substantiated this finding and cited the family's emotional history of dealing with illness as well as their false perceptions that lead to negative response to illness (Bogosian et al, 2009; Bostrom & Nilsagard, 2016; Falahi-Khoshknab et al, 2014).

The identification of family resources when a child is diagnosed with a chronic illness plays a role in a family coping and adapting to a chronic illness (McCubbin et al,

2002). Social determinants of health are defined as social and environmental factors such as social support, family income, community resources and access to health services (Secretary's Advisory Committee on National Health Promotion and Disease Prevention Objective for 2010, 2008). These factors can have a great impact on the health and well-being of families of children with chronic illness. Findings from this study suggested that most families of children with POMS who identified resources for coping better adjust and adapt to the diagnosis, yet some struggled with coping and adapting. Studies in the literature on adults with MS validated those families with identified resources improved quality of life because families have enhanced coping abilities (Diareme et al, 2006; Eliasova et al, 2014; Mulligan et al, 2016). Some of the caregivers in the study attributed their families' lacked resources due to the travel distance from the specialized center or from the fact that their primary treatment for their child was provided by someone who was less knowledgeable about POMS. These families openly talked about their limited resources. In some instances, caregivers reported that families did not have extra money to travel to specialized centers to receive education even once during a year. Other families who traveled >100 miles for pediatric MS had a perceived lack of resources because they did not know how to access the resources in their community, such as educational assistance for modifications in their child's school.

The caregivers of children with POMS also acknowledged the existence of family resources reported organizations like the National MS Society and other local organizations such vocational rehabilitation. The caregivers perceived that their families attained their knowledge of these resources from the center through informal resource education during the family's routine visits at the center.

Additional findings also suggested that families who had a powerful sense of religious beliefs coped and adapted to the diagnosis more efficiently. Although caregivers self-reported that all families were engaged in some type of religious affiliation, half of the caregivers identified their faith and religion as a source of strength, a resource that can also promote effective coping and adaptation. Those caregivers who discussed religion and God and their family's church life acknowledged that this resource assisted their families through what they would consider the toughest times of their lives after having a child diagnosed with POMS. Other caregivers talked about how their family turned to religion and prayer daily for comfort and strength to make coping with diagnosis achievable. Although there was very little information on the subject in the literature on families of adults with RRMS, the information further supported that religiosity plays a part in promoting adaptation (Ghafari et al., 2015).

Findings also suggested that families who had a dedicated support system have a greater ability to cope and adapt to the diagnosis. The caregivers in the study who lacked a support system exhibited signs of maladjustment, such as emotional turmoil, when the child with POMS was in a flare-up. Some of the caregivers reported things liked personally having a challenging time focusing on their jobs or the families' difficulty with prioritizing the other aspects of their lives like parenting or having a family social life. Some of the caregivers who lacked the support system not only struggled with dealing with the child during the crisis but they also struggled with dealing with the emotional needs of other family members and children. Some caregivers reported siblings having emotional outbursts and the family lacking the knowledge to deal with situation effectively. These findings were consistent with the literature regarding adults

with RRMS regarding the need for a support system to promote increased coping and adaptation in families of individuals with chronic illness (Koopman et al., 2006; Malcomson et al., 2008).

In addition to the identification of a dedicated support system, the acquisition of knowledge about the disease process can empower families of children with chronic illness to believe that they can meet the challenges that this disease may impose. Findings from this study suggested that families of children with POMS, diagnosed less than two years, struggle with adjustments due to lack of knowledge about disease management. Caregivers reported that families were overwhelmed with the amount of information that they received and that it was difficult of them to sort out and prioritize as a family. Families of children with POMS have a great deal of knowledge to acquire after the diagnosis has been made regarding but not limited to symptom management, acute and ongoing treatment regimen, and residual side effects. The severity of the disease and its manifestations influence the families' ability to cope with the amount of information needed to care for their children. In addition to the knowledge deficit, the first two years of the disease for a child with POMS may be unsettling due to increased disease activity. A review of the literature regarding families of adults with RRMS revealed that during the early phases of the disease a lack of knowledge about the disease led to anxiety, depression, family dysfunction and poor adjustment (K. Bjorgvinsdottir & S. Halldorsdottir, 2014; Ghafari et al., 2015). However, the literature did not address whether adaptation was affected by increased disease activity that could occur with POMS during the initial stages. These findings address a gap in literature regarding

families of children with POMS who may experience more issues with adapting to a diagnosis due to increased disease activity than families of adults diagnosed with RRMS.

Furthermore, the number of symptoms and the severity of the disease are variable among children with POMS. It is not surprising that findings from this study suggested that families of children with POMS who had unstable disease had greater difficulty with adjusting and adapting to the changes that come with the diagnosis. Caregivers of children with POMS who reported that their child with POMS had increased relapse rates, or increased burden of disease on their MRI and then required changes in medication had difficulty adjusting to the life changes associated with the diagnosis. Some caregivers also reported that children who have short periods between multiple relapses created a lot of anxiety and fear within some families because families had previous acquired knowledge that flare-ups are linked to disease progression. Those families had difficulty balancing the demands of the disease and this interfered with problem solving and the ability to cope because these families did not know what to expect. Families' fears related to the unpredictably of MS and progression of the disease despite disease modifying therapies (DMTs) resulted in maladjustment A review of literature on adults with RRMS does not address the issue of disease instability and its effects on coping.

In addition to other factors mentioned in the previous discussions, it is acknowledged that adjustments and adaptations of families of children with POMS may be dependent on many factors. Findings from this study indicated that the developmental level of the child with the disease influences the needs and concerns of families of children with POMS. This important finding is unique to this pediatric

population because children progress through varying stages of growth and development. The perceptions of the families of children with POMS' ability to cope may be dependent upon the developmental age and stage of growth and development at the time of diagnosis.

Caregivers in this study reported that most of the children (n=11; 65%) were diagnosed between the ages of 13 and 15 years of age. The rest were 16 and 17 years old (n=4; 20%) with only a few children who were less than ten years of age (n=2; 10%). For those children less than ten years of age, caregivers reported family concerns focused on the effects of MS on the growing child that may result in physical disability and cognitive effects on a child in this pre-pubescent stage. These caregivers knew that POMS might influence their child's ability to learn because of the possible cognitive effects of the disease during an optimal period of knowledge acquisition and maturation. In addition, caregivers also knew that POMS might cause physical impairments like difficulty walking.

Caregivers of children between the ages of 13 and 15 brought another perspective. Caregiver reported worries for this age group regarding how children with POMS progressed through puberty and were psychologically affected by having a chronic illness. Children have peaks in their growth and development and psychosocial desires to maintain relationships with peers. Caregiver and family concerns focused on their children's ability to keep up with their peers physically, socially, and academically, as well as finding balance and normalcy as a teenager. For the caregivers of children with POMS diagnosed between the ages of 16 and 18, additional family concerns arose. These caregivers reported that their family concerns were regarding their children's

future in college or seeking employment, and independence from the family to make transitions into adulthood.

In addition to these three age groups, the researcher gained insight into another unique population of caregivers of children who were diagnosed with POMS between ages 13 to 16 years and were ages 21 and 22 years at the time of the interview. Findings from this study suggested that caregivers of these young adults had similar concerns as the families of those children between 16 and 18 years of age. Additionally, the families were concerned about their children's future life goals such as having a family and the long-term outcomes of having MS as a child.

These findings regarding caregivers and family concerns, as it pertains to children with POMS who are growing and developing addresses a major gap in the literature. The physical, cognitive and psychological changes unique to the pediatric population with POMS may affect family adjustment, coping and adaptation. According the Resiliency Model, changes in the family life cycle, make families of children with chronic illness vulnerable because of the demands and changes that are required (McCubbin, Thompson & McCubbin, 1996). Some of these changes may cause stress and strain which may deplete interpersonal, social and economic resources and result in maladaptation or maladjustment (McCubbin et al., 1996). One of the caregivers shared her family's experience of having a child diagnosed with POMS at 16 who was age 21 at the interview and had progressive MS. This caregiver's family adaptation waxed and waned because of her child's disease progression. She shared her family's struggle to cope with their lives each day. The caregiver shared the acknowledgement of her family that her daughter would never be independent and have a true-life experience as her peers in the

initial stages of adulthood. Another one of the caregivers in this group shared her life experiences with having a child previously diagnosed with POMS who entered into motherhood. This caregiver shared an additional level of concern and anxiety for her child and her child's ability to care for her grandchildren. The caregivers of these young adults brought a unique experience to this study as families of child previously diagnosed with POMS who now have entered adulthood. This finding addresses a major gap in the literature, as this phenomenon has not been explored. There is a need for longitudinal research regarding young adults previously diagnosed with POMS who now have progressive and debilitating disease that may be dependent on their families for their physical and psychosocial needs.

In addition to family concerns regarding the developmental stages of the child diagnosed with POMS, consideration should be given to the needs of the siblings or other children living in the household with the child with POMS. Findings from this study suggest that changes in the family roles and responsibilities related to the diagnosis negatively affected some younger siblings of children with POMS. These caregivers reported that siblings displayed anger, jealousy and emotional outbursts when they felt the caregiver and other family members were more attentive to the needs of the child with POMS. Siblings displayed these emotions during times of acute illness, which left caregivers with feelings of inadequacy because they struggled with balancing the needs of the family with that of the child. The review of literature on adults with RRMS supported the fact that younger children whose parents have MS display a wide range of negative emotions due to their lack of understanding and their developmental level (Bostrom & Nilsagard, 2016 Pakenham, 2005). Findings from this study are consistent

with literature in that children exhibit a range of negative emotions when a close family member is dealing with a chronic illness (Bostrom & Nilsagard, 2016; Diareme et al., 2006; Paliokosta et al., 2009; Turpin, et al, 2008).

Although the caregivers reported that most of the siblings of children did not display any outward signs of negative emotions, some of those caregivers expressed their families' concerns regarding the imbalance they felt with parenting their other children. According to the Resiliency Model, this stressor can disrupt the family function and place a strain on their capabilities unless strategies are put into place to help families of children with POMS cope with the emotions of their other children (McCubbin, 1986). The review of literature on the families of adults diagnosed with RRMS support these findings in the POMS study that children are negatively impacted by the diagnosis of a MS (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016). However, the reason for the negative impact on children of adults with MS was due to symptoms that their parents were experiencing or the care that the child was required to provide for the parent (Bjorgvinsdottir & Halldorsdottir, 2014; Bostrom & Nilsagard, 2016). Additional findings from the study revealed that the lack of communication among families of children with POMS related to issues of adjustment and adaptation. Most of the caregivers reported that communication among their family members increased because of the diagnosis. Some of the caregivers who were having challenges with accepting the diagnosis reported that there was a lack of communication between the members of the family. One of the caregivers shared that their families had communication issues near the time of the initial diagnosis and that it took nearly two years to resolve them, which strained her relationship with her spouse and her spouse's relationship with their child

with POMS. One of the studies in the review of literature of families of adults with MS supported the findings that relationships can negatively be impacted if communication is lacking and result in conflict and opposition (Boland et al., 2012).

Furthermore, other caregivers expressed that communication among the family became difficult when the child was experiencing a flare-up or when the child was experiencing a new symptom and wanted to conceal it from the family. According to the Resiliency Model, affirming communication, which involves active listening and conversation to problem solve, is an effective way for families to adapt to a situation like a flare-up or the experiences of a symptom (McCubbin et al., 2001). If effective communication does not occur within families of children with POMS, the result may be ineffective problem-solving and ineffective coping when families are strategizing steps to take during a crisis like a relapse. The literature regarding families of adults living with RRMS supports the effectiveness of communication so that problems, expectations and needs can be addressed through in-depth discussion and collective problem solving (Bostrom & Nilsagard, 2016).

Finally, the analysis of the data revealed that balancing day-to-day family life and the achievement of adaptation is impacted by the ongoing health status of the child with POMS. POMS is a disease that waxes and wanes and has a great deal of variability among each family that it affects (MacAllister et al., 2007). When most of the caregivers reported how their families achieved balance, the caregivers discussed ways in which their families redefined their previous patterns of functioning to meet the needs of the child with POMS. Families reported that they modified family rituals like vacations and social outings to make adjustments for the child with POMS. The review of the literature

on families of adults living with RRMS is consistent with the finding in the study that MS is a "family matter, involving the recognition of each members' involvement in adapting to the diagnosis (Bostrom & Nilsagard, 2016).

Most of the caregivers referred or implied that their family life after the diagnosis of MS was the "new normal." Some made slight changes spending more time with each other and increasing communication among the family members and some made major changes like changing jobs and altered living arrangement. These caregivers also discussed how their families altered patterns of function as needed due to relapse or illness. They also discussed their families' resources of strength that included their extended family and friends, church family, and health professionals. These resources were identified as sources of support that were necessary to balance their lives. Despite the demand of POMS, they were coping with life from day to day. The families, who were adapting effectively, had gone through the necessary changes of adjustment. For some families, this period of adjustment was longer than for others.

Implications for Practice

The results of this study suggested that there are many areas of improvement needed in the care of families of children diagnosed with POMS. Healthcare providers must be proactive in alleviating any negative appraisal of the family due to misconceptions about the disease by providing initial and ongoing education regarding the disease in a timely manner. In addition, all families of children with POMS should be educated on the resources that are available in their community. Healthcare professionals must take into consideration the various stages of growth and development and how this

may impact how families cope with the diagnosis of POMS. The results of this study are important to healthcare professionals because this provides them with insight regarding the factors that influence the adaptation of families of children diagnosed with POMS.

The Resiliency Model can be used by health professionals in clinical practice as a tool to assist families in identifying their capabilities, personal and family values and resources as well as life challenges when there is a chronically ill child in the family (Walsh, 2003). In turn, the identification of these factors will assist health professionals to target key family processes to focus on to develop individual interventions that will enable optimal family functioning and well-being.

Families of children with POMS need ongoing education that promotes coping skills during times of disease instability to promote adjustment and adaptation during the disease process. Healthcare providers and other healthcare professionals need to take into consideration the need for assessing and making referrals to psychologists, social services and other professionals that are educated on assisting families regarding financial, social services, and developmental testing as well as other needs of the families including the siblings of those chronically ill children.

There is also a need for healthcare providers of children with POMS to develop a comprehensive education plan for families newly diagnosed with POMS. MS education tailored toward families of POMS should be made accessible using an online platform that may be viewed via cell phone or computer with the capability of being viewed at any time convenient for the person. The educational plan should include any pertinent information on topics such as treatment options, symptom management and research updates regarding POMS. Research on comprehensive educational programs for families

of children with chronic conditions has shown to have positive family effects on coping and managing chronic conditions more effectively and have promoted the family's involvement in self-management of chronic conditions (Hilliard, Powell & Anderson, 2016; Kieckhefer, Trahms, Churchill, Kratz, Uding & Villareale, 2013).

In addition, there is a need for clinical experts to development educational programs for general practitioners and adult neurologists providing care for children with POMS that address the unique experiences that are specific to the population. Literature on current practice has shown that patients often receive inadequate care, inappropriate disease management and little coordination when attempts to manage complex medical issues are handled by primary care physicians (Coleman, Mattke, Perrault, & Wagner, 2009). Proving an educational program for general practitioners and adult neurologists will allow for all children with POMS to receive competent care no matter the educational background of the provider. Healthcare providers must make referrals to social workers or counselors who can access the support system of the families so that individualized needs of the families can be determined. Local branches of the National MS Society can develop and provide families of children with POMS a list of community resources that can be utilized by families of children with POMS. Finally, a counseling service via telehealth is needed to educate families of children with POMS on coping and problem solving strategies related to managing POMS. These implications for practice will encourage families to become resilient despite the diagnosis of POMS.

Implications for Research

The findings from this study indicated the need for continued research to explore POMS from the perspective of families. Most family researchers agree that true qualitative family research is achieved through the in-depth comparison of multiple groups within a family to gather an accurate depiction of all persons living in the family and for the development of interventions that will impact the family (Gilgun & Sands, 2012). Future research with families of children diagnosed with multiple sclerosis should include comparison studies to gather different perspectives on the various relationships that exist within the context of the family. Another area that needs further exploration includes further research on the impact of POMS on siblings of children with POMS. This will provide insight into a population that may be directly impacted by the demands of chronic illness on the family rarely explored in the literature. Research on interventions involving the use of religiosity as an effective means of coping with POMS may be reasonable as well. Research regarding the long-term effects of this diagnosis on the family has not been conducted. Consequently, there is a need for longitudinal research on families of children who were previously diagnosed with POMS who have now entered young adulthood.

The findings in the study suggested that the role of religion and spirituality may have positive effects on the coping mechanisms of families. Further research is needed to explore the influence of spiritual and wellness counseling on the adaptation process of families of children with POMS.

Furthermore, the researcher acknowledges there were caregivers in this study that identified children with POMS that were considered atypical because these children did

not follow the natural trajectory of the disease or the children had additional issues along with the diagnosis of POMS. These atypical cases included: 1) a child diagnosed with POMS before the age of 4, 2) a teenager diagnosed at age 8 who later develops a co-morbid disease, 3) a young adult in early 20s diagnosed with POMS at 15 now with progressive debilitating disease, 4) a teen diagnosed with POMS that becomes pregnant within 6 months of her diagnosis and 5) a child diagnosed with POMS who has a parent diagnosed with MS. The identification of these atypical cases suggests the development of single or multiple case studies to depict how their cases may affect adaptation within the family.

Strengths and Limitations of the Study

As with all research, this qualitative descriptive study had strengths and limitations. This study had many strengths. The study was conducted by a researcher who was experienced and knowledgeable about the disease process of POMS and the effects that it has on those children diagnosed. The study was designed and conducted in a manner to ensure sound and credible research by the researcher adhering to the study protocols that were approved by the IRB. The methodology of one-on-one open-ended interviews provided rich and in-depth details of the families' experiences from the perspective of caregivers. In addition, the researcher had an established rapport with most of the participants prior to the research being conducted, which encouraged them to share their experiences without fear of judgement or bias.

Furthermore, the demographics regarding the children with POMS provided by the caregivers were representative of the children that were seen at the center for care of

POMS. The finding of this study will lead to the development of and educational interventions to educate families on effective coping strategies and identifying resources of support that will promote positive adaptation.

There were limitations to the study as well that need to be taken in consideration if this study is replicated in other populations. The interviews were conducted with the caregivers and the perspectives of the families discussed in this study may not be accurately represented. Participant recall of feelings and experiences may be inaccurate due to the lapse in time for those diagnosed for a longer length of time to the time of the interview. Because this research was conducted at a southeastern center, it may not pertain to families at other centers in other geographic locations. Furthermore, families at other geographic locations might be influenced by other cultural practices and beliefs, which may limit generalizability of the findings from this study. Although data saturation was met, and participants were representative of the center's population, it is unknown if the sample of participants were representative of the families who were maladapting or maladjusting. The nature of the researcher's established relationship may have influenced the report of findings. To minimize this risk, the researcher obtained informed consent and encouraged the participants to reflect on and describe their experiences. The researcher conducted a non-leading interview with the participants and maintained epochȇ, throughout the study.

Conclusion

This study sought to address gaps in the literature, as it was the first of its kind to examine the effects of POMS on families of children that are affected by the disease.

This contributed new knowledge about factors that influenced the adaptation of the families of children with POMS. Because this phenomenon had never been explored, healthcare professionals are provided insight regarding the day-to-day experiences of families of children with POMS and how they adjust and adapt to the diagnosis. The findings of the study suggest that future research should be conducted to build on the knowledge that was attained form the current study. Lastly, healthcare provider who provide care to these families must consider changes in practice that will promote positive coping, effective patterns of family functioning, and increased identification of personal and professional resources to aid families in adapting to a diagnosis of POMS.

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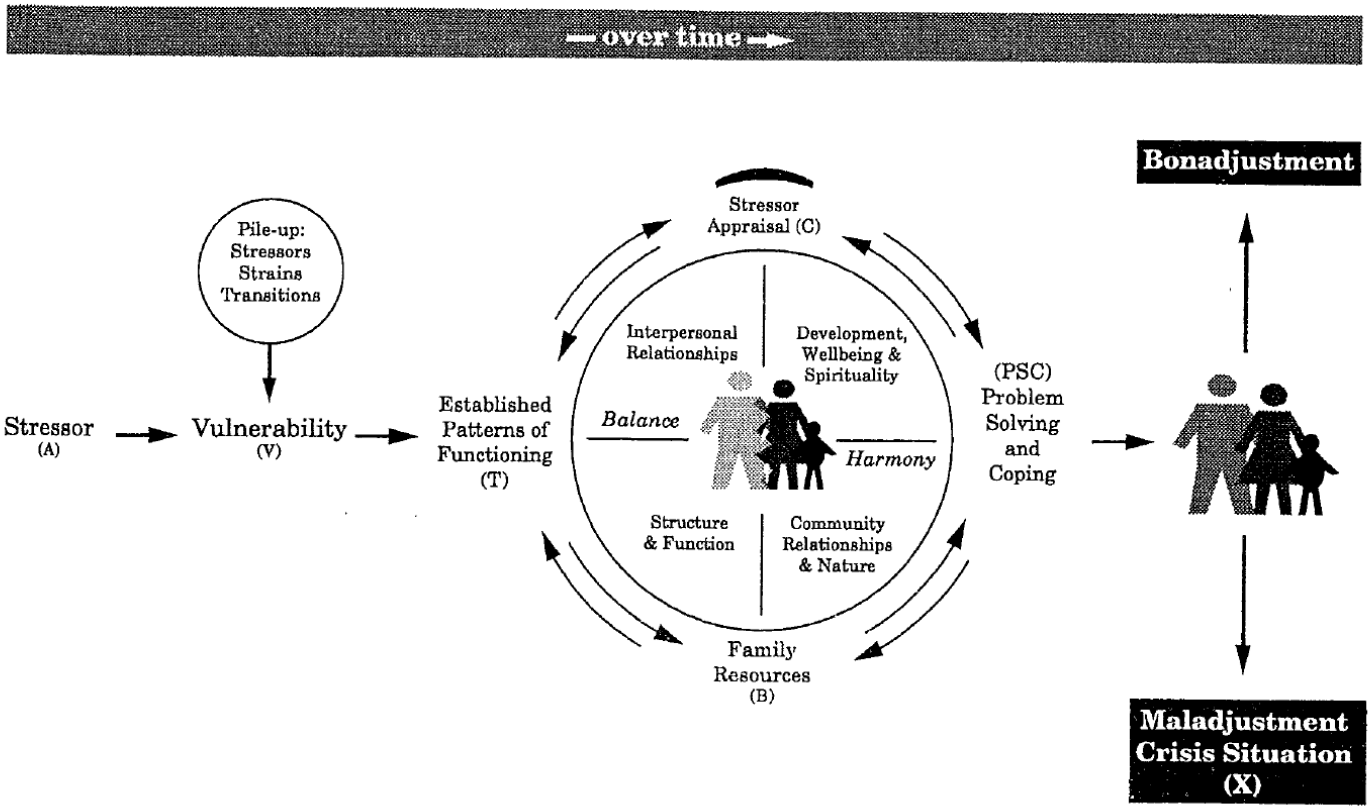
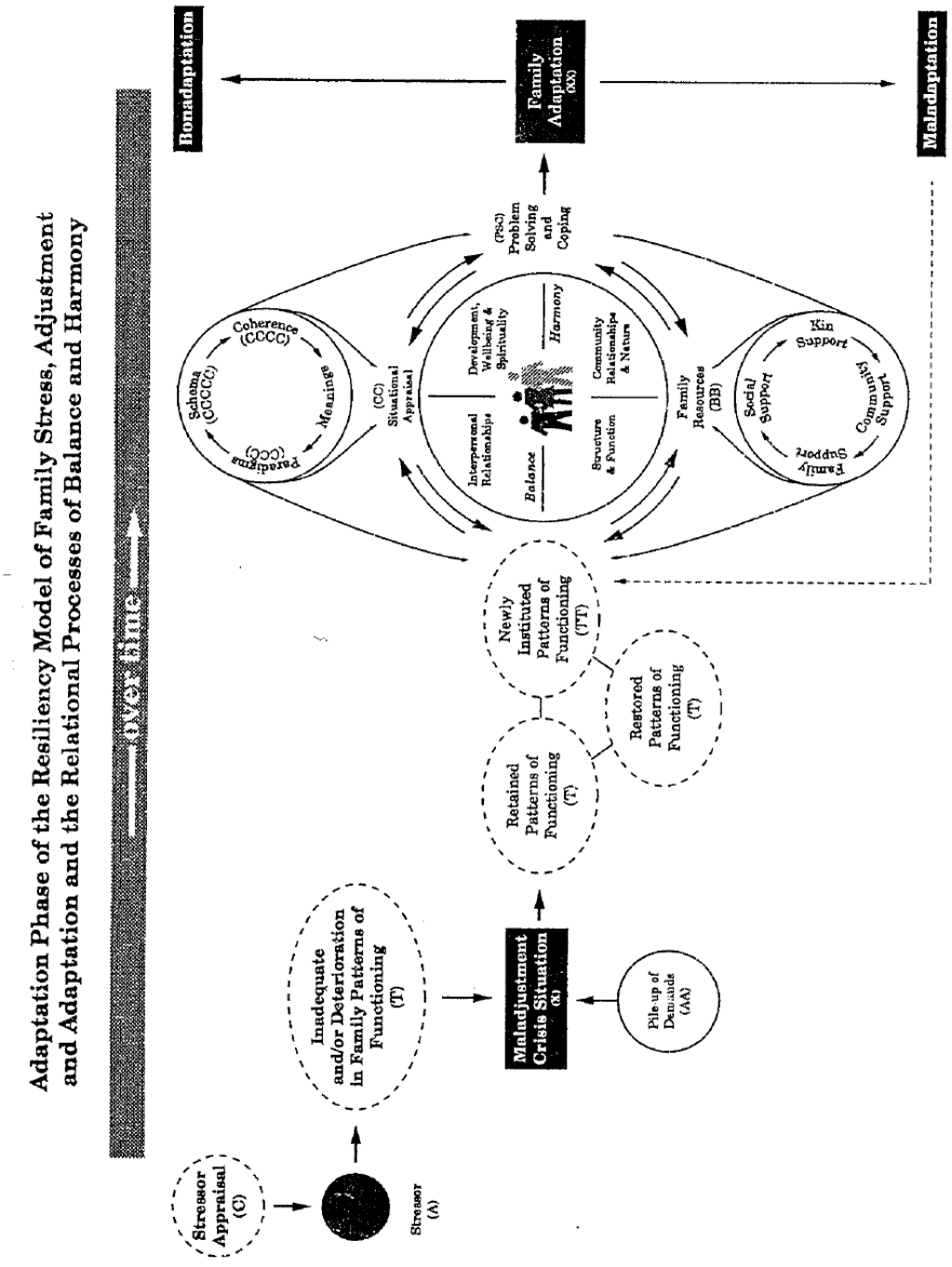
APPENDIX A

RESILIENCY MODEL

Adjustment Phase of the Resiliency Model of Family Stress, Adjustment and Adaptation and the Relational Processes of Balance and Harmony

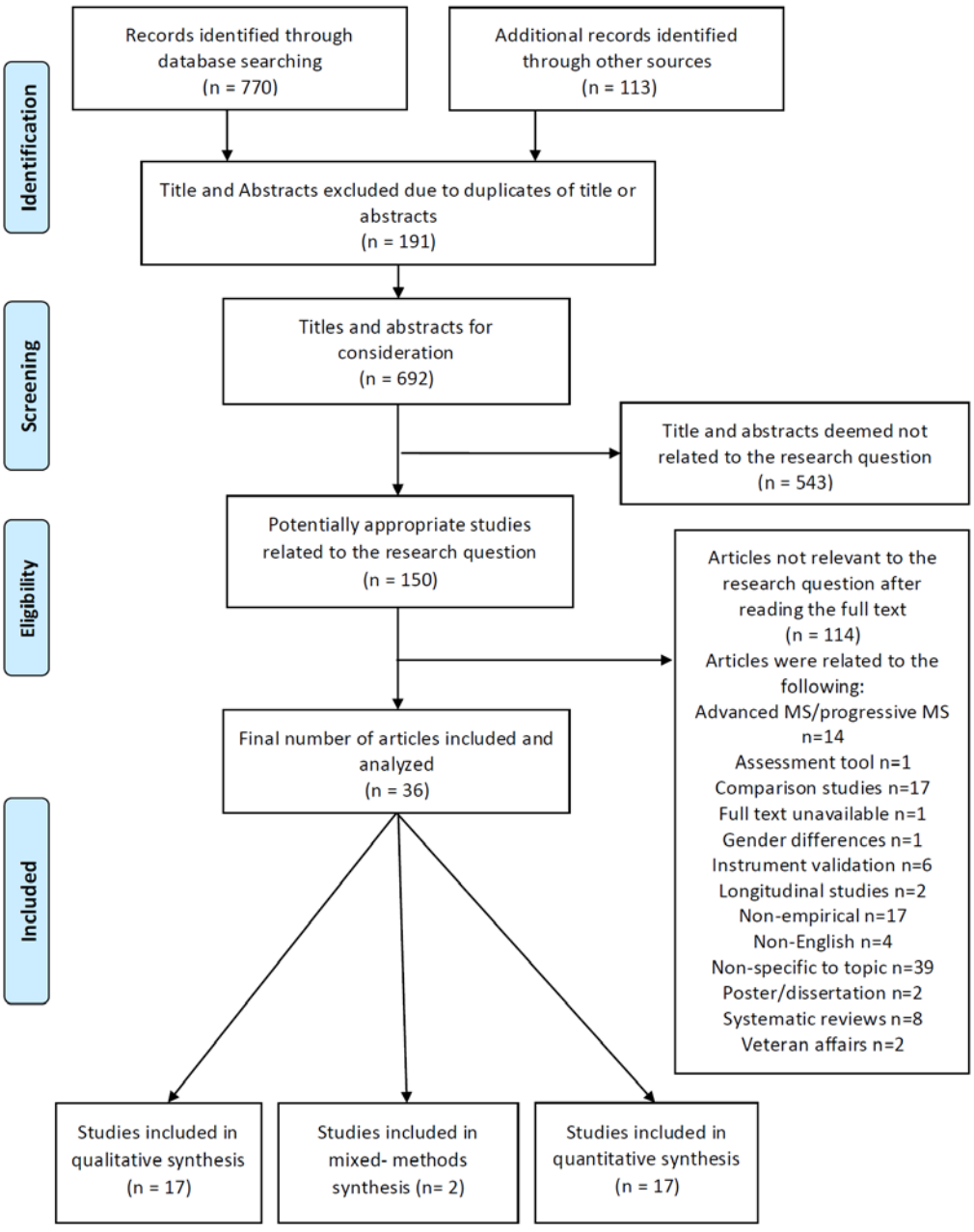



Adaptation Phase of the Resiliency Model of Family Stress, Adjustment and Adaptation and the Relational Processes of Balance and Harmony
Over Time
Stressor Appraisal (C)
Stressor (A)
Inadequate and/or Deterioration in Family Patterns of Functioning (T)
Maladjustment Crisis Situation (X)
Pile-up of Demands (AA)
Retained Patterns of Functioning (T)
Restored Patterns of Functioning (T)
Newly Instituted Patterns of Functioning (TT)
Bonadaptation
Family Adaptation (XX)
Maladaptation
(PSC) Problem Solving and Coping
(CC) Situational Appraisal
Schema (CCCC)
Coherence (CCCC)
Meanings
Paradigms (CCC)
Development, Well-being & Spirituality
Harmony
Community Relationships & Nature
Interpersonal Relationships
Balance
Structure & Function
Family Resources (BB)
Kin Support
Community Support
Social Support
Family Support

APPENDIX B

CONSORT FLOW DIAGRAM OF LITERATURE REVIEW

Consort Flow Diagram of Literature Review



APPENDIX C

FAMILY DEMANDS LITERATURE REVIEW MATRIX

Literature Review Matrix Regarding Family Demands in Adults with RRMS

Study (Year)	Design	Method	Sample	Findings Related to Demands
Aymerich et al., (2009)	Quantitative-Cross-sectional multicenter study of nine hospitals	Self-reported instruments-36 question Short Form Health Survey (patients); 12-item Short Health Survey (family caregivers) and short form General Health Questionnaire (psychological distress)	705 **individuals** (ages 16 to 73;77.8% with RRMS) and 551 caregivers	• Health related quality of life is lower in caregivers of people with MS, even with disease • Caring has a direct impact on psychological well-being
Bjorgvinsdottir & Halldorsdottir (2014)	Qualitative-thematic analysis	Unstructured interviews; 1-3 interviews per caregiver	11 **young caregivers** of individuals with MS (ages 5 to 18) when child began caring	• Young caregivers are "silent, invisible and unacknowledged" about the nature and consequences of their caregiver activities • Young caregivers received limited professional assistance • They had troubled memories, too many responsibilities • There was a lack of school specific support for children who are school aged
Bogosian et al., (2009)	Qualitative – inductive thematic analysis	Semi-structured telephone interviews	15 **partners** of individuals with MS (ages 32- to 59; 50% with RRMS)	• First few years were difficult as a partners because of difficulty adjusting due to the shock of the diagnosis and the unknown about the future • Sense of helplessness and loss of control • Feelings of social isolation because they felt they could not support and understand
Bostrom and Nilsagard , (2016)	Qualitative-content analysis	Focus group interviews with a semi-structure interview guide; interviews conducted with the	23 persons representing 10 **families** (the ill parent, the healthy	• Initial family reactions were colored by their experiences from first symptoms to MS diagnosis

		ill parent, the healthy parent and the child separately	parent and the child)	• Initial concerns of the ill parent were not related to themselves but to their children • Healthy parents neglected their own needs and expressed their feelings • Acute symptoms and unexplained symptoms created anxiety for children • Children worried that their parent would die from MS • Older children were very engaged and assistant parents • Younger siblings were angry and obstinate • Having a teenager in the family was stressful both on individuals and on the family
Diareme et al., (2006)	Quantitative-comparative analysis	Self -reported instruments; Child Behavioral Checklist (emotional and behavioral problems) Youth Self Report (children over 11); Beck Depression Inventory (parental depression and family dysfunction) Karnosky Perfor-mance Status Scale (illness related variables)	**Family** unit-56 MS patients, their spouses and a randomly selected child (ages 4 to 17)	• Children of parents with MS present with greater emotional and behavioral problems than their peers of similar age and gender whose parents have no physical illness especially when the ill parent is the mother • Internalizing problems in children of mothers with MS are predicted solely by the MS related impairment of the ill mother rather than any other variable examined • Mothers (especially ill mothers) were more depressed than their spouses
Fallahi-Khoshknab et al., (2014)	Qualitative-content analysis	Unstructured interview	25 **individuals** with MS (ages 20-55 years)	• Participants had a knowledge deficit, difficulty with concealing the disease due to misconceptions, fear or termination • Emotional reactions included anger, fear and anxiety, consternation, confusion and being demoralized

				• Family stressor was the result of their initial reaction to the diagnosis
Heward et al. , (2006)	Qualitative-grounded theory	In-depth semi-structured interviewing	9 **partners** of people with MS	• MS lead to provoked occupational constraints for the partner of individuals with MS • MS inspired occupational opportunities
Koopman et al; (2006)	Quantitative	Questionnaire Was developed from 4 focus groups	**Dyad**-353 individuals with MS and 240 significant others	• Top 10 Needs of the MS patient and significant others are: 1. To have the support of family and friends 2. To know doctors are interested 3. To have a supportive family doctor 4. To feel productive in life 5. To receive regular newsletters from the MS clinic 6. To be encourages to maintain control of my life 7. To receive newsletters from the MS Society 8. To know that the individual MS team members that I see are interested 9. To hear information about the future as it relates to how my condition is now 10. To know that concerns have been heard by the MS clinic
Labiano - Foutcuberta et al., (2015)	Quantitative	Structured interview with individuals with MS (demographic data, comorbidity, medications, age of MS onset and disease evolution), clinical, neurological exam, psychiatric assessment (EDSS, cognitive testing caregivers given a questionnaire	63 **individuals** with MS (ages 37-57) and their 63 corresponding **caregivers**, 59 **matched controls**	• Information processing speed impairment is independently associated with more severe depressive symptoms of caregivers of MS patients, reflecting a further deterioration of family

		regarding demographic care caregiving related data as well as self- reported instrument (Beck Anxiety Inventory and Beck Depression Inventory		
Malcomson, et al., (2008)	Qualitative – thematic analysis	2 Focus groups	13 **individuals** with MS	• MS patients learned something was wrong • They received a name to the diagnosis • They experienced a lack of professional support • They longed for unchanging family relationships • They had to have adjustments to employment circumstances and social life • They identified challenges
Pakenham et al., 2012	Qualitative thematic analysis	2 open ended questions in a questionnaire survey	119 **individuals** with MS and 64 **partners**	• Parenting difficulties themes emerged and included: shortness of time, activities, implications of MS on the family, fatigue related to day to day difficulty, sons and the patient's inability to participate in their activities, issues with partners, trouble with keeping up with daughters, difficulty with housework, family (represented parents with MS) and mood change in the parent with MS
Steck et al., (2007)	Quantitative	Self-reported questionnaires were completed by the parents; demographic were used to measure illness status and illness severity; Beck Depression Inventory (parent	144 **families** (144 individuals with MS, 109 partners and 192 children)	• 59% of all patients and 20% of the partners evaluate themselves as being depressed • Psychological distress affects not only the chronically ill patient but all the caregivers • Severe disease is associated with

		depression); Child Behavior Checklist (child mental health and behavior completed by parent); Youth Self Report (completed by children- behavioral and psychological status)		depression of ill and healthy parents • Both parents and offspring reported significantly higher scores for internalizing disorders • The higher the depression score of ill mothers and even more of healthy parents, the higher the psychosocial problems are evaluated in their children
Turpin et al., (2008)	Qualitative – thematic analysis	Semi-structured interviews	8 **children** (ages 7 to 14) whose parent was diagnosed with MS	• Three major themes developed related to the day-to-day experiences of the participants 1. Changing roles and responsibilities restricted their developmentally appropriate occupations 2. Emotional Impact- participants experienced conflicting emotions 3. Things that helped depended on the nature of the stressor

APPENDIX D

FAMILY RESOURCES LITERATURE REVIEW MATRIX

Literature Review Matrix Regarding Family Resources in Adults with RRMS

Study (Year)	Design	Method	Sample	Findings Related to Demands
**Diareme et al., (2006)	Quantitative-comparative analysis	Self -reported instruments; Child Behavioral Checklist (emotional and behavioral problems) Youth Self Report (children over 11); Beck Depression Inventory (parental depression and family dysfunction) Karnosky Performance Status Scale (illness related variables)	**Family** unit-56 MS patients, their spouses and a randomly selected child (ages 4 to 17)	Good family functioning is a protective factor against the development of delinquent behaviors and other aggressive manifestations in the children of mothers with MS
Eliasova et al., (2014)	Quantitative-comparative analysis	WHOQOL-BREF (short version)-evaluation of general quality of life and 4 quality of life domains	91 **individuals** with MS (46 attended a self-help group and 35 did not)	The self-help group has better scores related to quality of life in all three domains: 1) physical (sleep quality and sexual satisfaction),2)surviving, and 3)social relations (satisfaction in personal relationships , economic circumstances and self-contentment and coping with negative feelings
Ghafari et al., (2014)	Qualitative content analysis	Unstructured interviews	25 **individuals** with MS (ages 20-55)	Hopes was a source for couples; physical support was provided (physical support, physical care, financial support
Horton et al., (2015)	Qualitative Content analysis	In-depth semi-structured interviews	**Dyad**- 5 individuals with MS and their spouses	Three themes emerged from the analysis: 1. Maintaining independence

				2. Overcoming isolation 3. Negotiating if an intervention such as exercise is worth it
Mulligan et al., (2016)	Qualitative content analysis	Semi-structured telephone interviews at 1 month and 3 months	23 **individuals** with MS (ages 37 to 63; 61% RRMS)	Two themes emerged from the analysis: 1. Achieving behavior change to manage fatigue (reflective learning, taking control and developing new habits 2. Whole of life effects (building resilience, a new support network, obtaining balance and improved family dynamics
**Pakenham et al., (2012)	Qualitative thematic analysis	2 open ended questions in a questionnaire survey	**Dyad**-119 individuals with MS and 64 partners	Family resources included school, time, assistance, chores, friends, family, spouses, sons and driving
** Indicates the study was evaluated in the discussion of literature of another factor regarding adaptation.				

APPENDIX E

FAMILY APPRAISAL LITERATURE REVIEW MATRIX

Literature Review Matrix Regarding Family Appraisal in Adults with RRMS

Study (Year)	Design	Method	Sample	Findings Related to Demands
**Bjorgvinsdottir & Halldorsdottir (2014)	Qualitative-thematic analysis	Unstructured interviews; 1-3 interviews per caregiver	11 **young caregivers** of individuals with MS (ages 5 to 18) when child began caring	Caregivers appraise themselves as "silent, invisible and unacknowledged" about the nature and consequences of their caregiving activities
**Bogosian et al., (2009)	Qualitative – inductive thematic analysis	Semi-structured telephone interviews;	15 **partners** of individuals with MS (ages 32-59)-50% with RRMS	Providing care to a person with MS can have a negative impact on the social life of caregivers
**Bostrom and Nilsagard , (2016)	Qualitative-content analysis	Focus group interviews with a semi-structure interview guide; interviews conducted with the ill parent, the healthy parent and the child separately	23 persons representing 10 **families** (the ill parent, the healthy parent and the child	Both their individual history and their family history shaped how they looked upon and reacted to the situation To be unable to manage tasks without support gives rise to different feelings and accepting being a person in need or help was perceived as difficult
**Fallahi-Khoshknab et al., (2014)	Qualitative-content analysis	Unstructured interview	25 **individuals** with MS (20-55 years of age)	Most participants had false perceptions about MS. They had inadequate information and generally negative public attitudes toward MS.
Liedstrom et al., (2010)	Mixed Methods	Interview followed by a questionnaire about QOL	44 **next of kin** (29 spouses/cohabiting partners, 10 parents, 3 siblings, 2 adult children)	Most of the next of kin indicated a trusting and secure relationship with the cohabitating partner, but others described a strained situation with an unsatisfactory married/cohabiting life The next of kin spoke of a decrease in freedom, self-actualization and security also for a more negative general mood and negative emotional experience The next of kin considered that QOL also had to do

				with being healthy, having freedom and doing what they wanted to do. The next of kin in this study spoke of having learned to see life in another way They no strove for material things or opportunities for travel

APPENDIX F

FAMILY PROBLEM SOLVING AND COMMUNICATION LITERATURE REVIEW MATRIX

Literature Review Matrix Regarding Family Problem Solving and Communication

Study (Year)	Design	Method	Sample	Findings Related to Demands
**Bogosian et al., (2009)	Qualitative – inductive thematic analysis	Semi-structured telephone interviews	15 **partners** of individuals with MS (ages 32 to 59; 50% with RRMS)	• Most participants tried to find out more information about the illness and possible treatment, accepted new challenges and implemented solutions to their practical problems as they came along • Spousal attempt to overcome the constant and daily inconvenience in their lives by strategies such as taking each day as it comes and maneuvering around the present challenge
Boland et al., (2012)	Qualitative-thematic analysis interpretative phenomenology	Semi-structured interviews	7 **Dyads** (7 individuals with MS and 7 spouses	• The overarching theme of coping together on a daily basis was described as “peaks and troughs” • If the coping styles were oppositional, there was an increased likelihood of conflict and frustration • Shared responsibility in domestic life normalizes their situation and gave both something to focus on sharing a sense of pride • The couples perceived themselves as being a unit and that this sense of togetherness gave them a sense of purpose when their emotional stability became threatened
**Bostrom and Nilsagard , (2016)	Qualitative-content analysis	Focus group interviews with a semi-structure interview guide; interviews conducted with the ill parent, the	23 persons representing 10 **families** (the ill parent, the healthy parent and the child)	• Family coping was achieved through open communication; kids were included; some people involved others right from the time of diagnosis, while others took it

		healthy parent and the child separately		bit by bit over time; some grew with the situation • Some families talked about retrogressive behavior in the adolescent-they did not show concern about MS but reacted strongly to small setbacks • The families had to have an ongoing negotiation about division of responsibility
Ehrensperger et al., (2008)	Quantitative	Semi-structured index with the parental couple and each child was used to derive a coping index using statistical analysis; patients were assess using the following: 1) a comprehensive battery of neuropsychological tests, 2) neuro-logical status (Kurtzke Expanded Disability Status Scale), 3) depressive symptoms (Beck Depression Inventory)	44 **families** with a parent suffering from MS, their partners and offspring	• A high coping index of the ill parent indicates successful coping with the disease and is associated with few depressive symptoms • The intact ability to simultaneously pay attention to different stimuli and lower estimated pre-morbid verbal intelligence • Coping behavior in this study was not a result of the degree of physical impairment nor the course of the disease • Lower levels of depression were related to positive coping strategies; higher levels of depression were associated with negative coping strategies.
**Fallahi-Khoshknab et al., (2014)	Qualitative-content analysis	Unstructured interview	25 **individuals** with MS (ages 20 to 55 years)	• The patient used religiosity to cope with the disease: their faith in God, and their resort to prayer
**Ghafari et al., (2014)	Qualitative content analysis	Unstructured interviews	25 **individuals** with MS (ages 20 to 55)	• Emotional support from their partners is more effective than their physical help in coping with the disease

**Malcomson, et al., (2008)	Qualitative – thematic analysis	2 Focus groups	13 **individuals** with MS (ages 40-67)	• Successful coping includes: proactivity; perspective and control (self-management); advice to others and recommendations as to how services could be improved and developed
Mauseth & Hjalmhult (2016)	Qualitative-Grounded theory-open line coding	Semi-structured interviews	15 **adolescents** (ages 12-18) who had a parent diagnosed with MS	• Adolescents balanced their needs by: 1) reflecting, 2) adjusting; 3) taking responsibility, and 4)seeking respite
Mikula et al., (2014)	Mixed methods–cross sectional	Semi-structured interview and a neurological exam as well as medical records and EDSS; health related quality of life-36 item short form; 26 item coping self-efficacy scale	113 **individuals** with MS	• Stopping unpleasant emotions and thoughts seems to be the most important type of coping in MS patients
Paliokosta et al., (2009)	Quantitative-multi-site	Semi-structured interview (Socio demographic data, child developmental history and history of parental illness; self-reported measures: Child Behavioral Checklist (completed by parents regarding children); Beck Depression Inventory (depression in parents); Family Assessment Device (family dysfunction); Karnofsky Performance Status Scale (patient functional impairment classification)	56 **families (**a parent with MS, well parent and a randomly selected child (ages 4-17)	• There was a significant correlation between the age of children and communication around illness • older children had significantly more chances to have some or full information about parental illness than younger children • Communication around parental MS in the family could be a contributing factor for psychosocial problems of children

**Turpin, et al., (2008)	Qualitative Inductive thematic analysis	Semi-structured interview	8 **children** of individuals with MS	• Coping strategies employed by the children depended on the nature of the stressor and if it could be changed • They coped by expressing emotions, changing their own behavior in response to their demands placed on them • Some children used social supports (parents, teachers and friends) as a method of coping • Older children who were more competent and who has more personal freedom, developed their own interest and social relationships as a way of coping
** Indicates the study was evaluated in the discussion of literature of another factor regarding adaptation.				

APPENDIX G

FAMILY ADAPTATION LITERATURE REVIEW MATRIX

Literature Review Matrix Regarding Family Adaptation in Adults with RRMS

Study (Year)	Design	Method	Sample	Findings Related to Demands
**Bostrom and Nilsagard , (2016)	Qualitative-content analysis	Focus group interviews with a semi-structure interview guide; interviews conducted with the ill parent, the healthy parent and the child separately	23 persons representing 10 **families** (the ill parent, the healthy parent and the child)	• Persons with MS require more time to adjust and come to terms with the diagnosis.
Ghafari et al., (2015)	Qualitative content analysis	Unstructured interviews	25 **individuals** with MS (ages 20-55)	• 7 Themes and 18 sub-themes emerged regarding adaptive strategies: 1. Religiosity 2. Information seeking 3. Seeking support 4. Maintaining hope 5. Concealing the disease 6. Emotional reaction 7. Fighting the disease and disability
Hwang, et al., (2011)	Quantitative-correlational	Self-reported measures: Leeds Multiple Sclerosis Quality of Life Scale (quality of life); 18-item questionnaire that included 3 MS adaptation scales (adjusted self-concept, social support and accessibility	66 **individuals** with MS (ages 18-71)	• Adjusted self-concept, social support and accessibility is critical in the psychological adaptation to the course of MS
Starks et al., (2010)	Mixed Methods	Questionnaires and semi-structured interviews	8 **Dyad** couples	• Couples were placed in two groups based on their patterns of adaptation: “being in sync” or “being out of sync” • Couples that were ‘in sync” with each other had compatible styles and world view. They were able to respond together to problem solve

				challenges posed by MS • The couples that were out of sync who adapted and adjusted to change at a different pace and used strategies that focused on different goals or priorities which made it difficult for them to work together
** Indicates the study was evaluated in the discussion of literature of another factor regarding adaptation.				

APPENDIX H

SCREENING TOOL FOR ADAPTSPOMS STUDY

Screening Tool for ADAPT2POMS Study

Name of Potential Participant(s)	1. 2.
Name of Child with MS	
Home Phone Number	
Cell Phone Number	
Mailing Address	Street Address:
	City, State and Zip
Was the mailing address confirmed via phone call?	○ yes ○no

1. The child of the participant is between the ages of 0 months and 23 years	Y/N
2. The child of the participant has been diagnosed with clinical definite MS for least 6 months	Y/N
3. The potential participant is a caregiver of the child with POMS	Y/N
4. Does the child with POMS live in the household with the participant most of the time?	Y/N
5. The participant's primary language is English	Y/N
6. The participant is able to read the English language	Y/N
7. The participant is at least 18 years of age	Y/N

APPENDIX I

RESEARCH FLYER

ADAPT2POMS Study:
Adapting To Pediatric Onset Multiple Sclerosis

Exploring Family Factors that Influence the Adaptation of POMS

- **What is the purpose of this study?**

  The purpose of this research study is to identify family factors that influence a family's adaptation to having a child with pediatric onset multiple sclerosis (POMS).

- **Who can participate in this study?**
  - A primary caregiver of a child who has been diagnosed with clinically definite MS for at least 6 months
  - A person that is 18 years of age or older and is able to speak and read English
  - A person who has a child with POMS that has received previous or on-going care and consultation in the last 2 years at the Center for Pediatric Onset Demyelinating Disease
  - A person who was the caregiver of a child with POMS that received previous or on-going care and consultation at the Center for Pediatric Onset Demyelinating Disease but the young adult has now transitioned from the pediatric center to the UAB Adult MS Clinic

- **If I agree to the study, what will I have to do?**

  If you agree, you will have to participant in a 60 minute telephone interview or an in-person interview at your convenience. You will also have to fill out a demographic form and small survey on family hardiness.

- **How are my rights and confidentiality protected?**

  Before you take part in the study, you will receive detailed information about the study. You will be asked to sign a consent form verifying that all your questions have been answered to your satisfaction. Only members of the research team will know your identity. You have the right to withdraw from the research at any time during the research without compromise to your child's care or penalty to you.

- **What are the benefits of participating in the study?**

  Gratification in being able to talk to others about their family's personal experiences. Satisfaction that their involvement may someday be used to help develop programs for other families to develop coping skills and other effective strategies when faced with a POMS diagnosis. Participants are compensated for their time that they spend in completing the study.

- **What are the risks of participating in the study?**

  Discussing your experiences may cause minimal emotional distress. If the distress is too overwhelming the interview will be stopped.

TO ENROLL IN THE STUDY OR OBTAIN MORE INFORMATION CONTACT

Yolanda Harris—205-934-0639 email: yoharris@uab.edu

Yolanda Harris, MSN, CPNP-AC, MSCN
Primary Investigator
UAB School of Nursing

Version 2 05-08-2017

APPENDIX J

LETTERS OF SUPPORT

June 12, 2017

RE: Letter of Support for ADAPT2POMS Study

To [Name of Potential Participant]:

This letter is to confirm my enthusiasm and strong support for Yolanda Harris to complete her PhD dissertation research "**A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers".** This study will involve caregivers of children who have been diagnosed with pediatric onset multiple sclerosis at the University of Alabama at Birmingham Center for Pediatric Onset Demyelinating Disease (CPODD).

I am very happy that she is working toward the completion of this goal that she set nearly a decade ago. At that time, she began to collaborate with me and the multi-disciplinary team at the inception of the center. Her research will expand the knowledge regarding how families are impacted the diagnosis of children with multiple sclerosis. It is our hope that this study will lead the way to the development of much needed family interventions that will help other families to cope and adapt to the diagnosis of multiple sclerosis.

As you know, I am the Director of CPODD at Children's of Alabama. We see pediatric MS patients from across the Southeast. I do not foresee Yolanda will not have any problems enrolling families of patients into this study as we have a substantial pediatric population.

I look forward to our continued collaboration.

Sincerely,

Jayne Ness

Dr. Jayne M. Ness, MD, PhD
Associate Professor, UAB Department of Pediatrics
Division of Pediatric Neurology
Director, Center for Pediatric Onset Demyelinating Disease

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UAB SCHOOL OF MEDICINE

Knowledge that will change your world

Department Of Neurology

May 7, 2017

UAB Institutional Review Board
Administration Building(AB)
Room 470
701 20th Street South, Birmingham, AL 35294-0104

I am writing on behalf of the University of Alabama at Birmingham Division of Neuroimmunology and Multiple Sclerosis (MS) and the Multiple Sclerosis Center which now includes a collaboration between the adult UAB MS Clinic and the Center for Pediatric Onset Demyelinating Disease (CPODD) in the UAB Department of Pediatric Neurology at Children's of Alabama. I currently serve as the co-director of the adult UAB MS Clinic and the co-director of CPODD.

A great majority of young adult patients diagnosed with pediatric onset MS transition from CPODD to the adult UAB MS Clinic. These young adults are seen for routine care in my clinic. I am in support of Yolanda Harris expanding her recruitment efforts for her dissertation proposal to include family caregivers of young adults who carry a pediatric onset MS diagnosis. She has built rapport with these families and they will understand the importance of this research project as well as those she will develop in the future. Furthermore, this population will yield a diverse group of participants and families that effected by pediatric multiple sclerosis in the Southeast and will enhance her research findings.

Sincerely,

K. Bashir

Khurram Bashir, M.D., MPH
Professor and Vice Chair for Education, Department of Neurology
Director, Division of Neuroimmunology and Multiple Sclerosis
Director, Neurology Residency Training Program

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APPENDIX K

PHONE SCRIPT FOR RECRUITMENT

Phone Script for Recruitment for ADAPT2POMS Study

*To be utilized when potential participants are scheduled to come to a clinic visit in the upcoming week and are contacted phone the following conversation will be addressed with each person.

PI or research staff: *"**Hello, potential participant's name. My name is caller's name. I am on staff at the Pediatric MS where your child has an upcoming appointment with Dr. Ness and her staff. I am calling to let you know about a research study that you may be potentially interested in at the Pediatric MS Center. It is called the ADAPT2POMS or the Adaptation to Pediatric Onset MS study. Do you have a moment to talk with me about this?"***

Wait for a response from the potential participant.

PI or research staff: ***"This study is to explore how families adapt to a diagnosis of pediatric onset multiple sclerosis. It has three components to it. One is component is a 60 minute, one-on-one interview with the person conducting the research, which happens to be the nurse practitioner in the clinic. The interview can be done over the phone or in person and if you chose to participate you can choose the way that is convenient for you. The questions in the interview will talk about what changes have been made within your family since one of your children has been diagnosed with MS. The interview will be audio taped and will be conducted in a private room without distraction.***

Do you have any questions about the interview?

Wait for a response from the potential participant.

PI or research staff: "***There is one other form that has to be filled out to complete the project. It is a demographic form that asks you about work, home, your child with MS and other children and adults living in your home. It will take about 15 to 20 minutes to fill out.***

PI or Research staff: ***"Do you have any questions so far?"***

Wait for a response from the potential participant.

PI or Research staff: "***OK. Let me tell you more. The participant has to be a primary caregiver that is at least 18 years or older. A primary caregiver is the person that provides care for the child most of the time at home. The participant also has to be able to speak and read English. The child of the participant has to be between the ages of 0 and 23 years of age and must have been seen in the last year. The child of the participant must have been diagnosed with clinically definite MS for at least six months."***

"***Do you meet those qualifications?"***

Wait for a response from the potential participant.

If the answer is yes, then the PI or Research staff will answer the following:

"Would you be interested in participation with this study?"

If yes, they are willing to participate, the PI will set up a time/date for informed consent and an interview or the staff will inform the potential participants that the PI will call to set up a time/date for informed consent and the interview.

If no, the researcher or staff will thank them for their call and express to them to please feel free to contact the researcher again for any questions about the study.

APPENDIX L

IRB APPROVAL OF INFORMED CONSENT AND AUTHORIZATION

THE UNIVERSITY OF ALABAMA AT BIRMINGHAM

Institutional Review Board for Human Use

Form 4: IRB Approval Form
Identification and Certification of Research
Projects Involving Human Subjects

UAB's Institutional Review Boards for Human Use (IRBs) have an approved Federalwide Assurance with the Office for Human Research Protections (OHRP). The Assurance number is FWA00005960 and it expires on November 8, 2021. The UAB IRBs are also in compliance with 21 CFR Parts 50 and 56.

Principal Investigator:	HARRIS, YOLANDA
Co-Investigator(s):	
Protocol Number:	**X161103004**
Protocol Title:	*A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis form the Perspective of Caregivers*

The IRB reviewed and approved the above named project on 12/8/16. The review was conducted in accordance with UAB's Assurance of Compliance approved by the Department of Health and Human Services. This Project will be subject to Annual continuing review as provided in that Assurance.

This project received EXPEDITED review.

IRB Approval Date: 12/8/16

Date IRB Approval Issued: 12/8/16

IRB Approval No Longer Valid On: 12/8/17

HIPAA Waiver Approved?: Yes

Partial HIPAA Waiver Approved?: Yes

Expedited Reviewer
Member - Institutional Review Board
for Human Use (IRB)

Investigators please note:

The IRB approved consent form used in the study must contain the IRB approval date and expiration date.

IRB approval is given for one year unless otherwise noted. For projects subject to annual review research activities may not continue past the one year anniversary of the IRB approval date.

Any modifications in the study methodology, protocol and/or consent form must be submitted for review and approval to the IRB prior to implementation.

Adverse Events and/or unanticipated risks to subjects or others at UAB or other participating institutions must be reported promptly to the IRB.

470 Administration Building
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irb@uab.edu

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THE UNIVERSITY OF ALABAMA AT BIRMINGHAM

Institutional Review Board for Human Use

PI: HARRIS, YOLANDA
Protocol # X161103004

UAB IRB Approval of Waiver of Informed Consent and/or Waiver of Patient Authorization

☐ **Approval of Waiver of Informed Consent to Participate in Research.** The IRB reviewed the proposed research and granted the request for waiver of informed consent to participate in research, based on the following findings:

1. The research involves no more than minimal risk to the subjects.
2. The research cannot practicably be carried out without the waiver.
3. The waiver will not adversely affect the rights and welfare of the subjects.
4. When appropriate, the subjects will be provided with additional pertinent information after participation.

Check one:
- ☐ **and** Waiver of Authorization (below)
- ☑ **or** Waiver of Authorization (below)
- ☐ Waiver of Authorization not applicable

☑ **Approval of Waiver of Patient Authorization to Use PHI in Research.** The IRB reviewed the proposed research and granted the request for waiver of patient authorization to use PHI in research, based on the following findings:

1. The use/disclosure of PHI involves no more than minimal risk to the privacy of individuals
   i. There is an adequate plan to protect the identifiers from improper use and disclosure.
   ii. There is an adequate plan to destroy the identifiers at the earliest opportunity consistent with conduct of the research, unless there is a health or research justification for retaining the identifiers or such retention that is otherwise required by law.
   iii. There is an assurance that the PHI will not be reused or disclosed to any other person or entity, except as required by law, for authorized oversight of the research study, or for other research for which the use or disclosure of PHI would be permitted.
2. The research cannot practicably be conducted without the waiver or alteration.
3. The research cannot practicably be conducted without access to and use of the PHI.

—OR—

☐ **Full Review**

The IRB reviewed the proposed research at a **convened meeting** at which a majority of the IRB was present, including one member who is not affiliated with any entity conducting or sponsoring the research, and not related to any person who is affiliated with any of such entities. The waiver of authorization was approved by the majority of the IRB members present at the meeting.

Date of Meeting

Signature of Chair, Vice-Chair or Designee

Date

☑ **Expedited Review**

The IRB used an **expedited review procedure** because the research involves no more than minimal risk to the privacy of the individuals who are the subject of the PHI for which use or disclosure is being sought. The review and approval of the waiver of authorization were carried out by the Chair of the IRB, or by one of the Vice-Chairs of the IRB as designated by the Chair of the IRB.

12/8/16
Date of Expedited Review

Signature of Chair, Vice-Chair or Designee

Date 12/8/16

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THE UNIVERSITY OF ALABAMA AT BIRMINGHAM

Institutional Review Board for Human Use

PI: HARRIS, YOLANDA
Protocol # X161103004

UAB IRB Approval of
Partial Waiver of HIPAA Authorization
to Use PHI in Screening for Research

☑ **Patient Authorization: Approval of Partial HIPAA Waiver to Use PHI in Screening for Research.** The IRB reviewed the proposed research and granted the request for a "partial HIPAA waiver," to allow the proposed use of protected health information (PHI) in screening for research, based on the following findings:

1. The use/disclosure of PHI to screen candidates for research involves no more than minimal risk to the privacy of individuals
   a. There is an adequate plan to protect the identifiers from improper use and disclosure.
   b. There is an adequate plan to destroy the identifiers at the earliest opportunity consistent with conduct of the research, unless there is a health or research justification for retaining the identifiers or such retention is otherwise required by law.
   c. The PHI will not be reused or disclosed to any other person or entity, except as required by law, for authorized oversight of the research study, or for other research for which the use or disclosure of PHI would be permitted.
2. The screening cannot practicably be conducted without the waiver or alteration.
3. The screening cannot practicably be conducted without access to and use of the PHI.

—OR—

☐ **Full Review**
The IRB reviewed the proposed research at a **convened meeting** at which a majority of the IRB was present, including one member who is not affiliated with any entity conducting or sponsoring the research, and not related to any person who is affiliated with any of such entities. The partial waiver of authorization for screening was approved by the majority of the IRB members present at the meeting.

_____
Date of Meeting

_____
Signature of Chair, Vice-Chair or Designee

_____
Date

☑ **Expedited Review**
The IRB used an **expedited review procedure** because the research involves no more than minimal risk to the privacy of the individuals who are the subject of the PHI for which use or disclosure is being sought. The review and approval of the partial waiver of authorization for screening was carried out by the Chair of the IRB, or by one of the Vice-Chairs of the IRB as designated by the Chair of the IRB.

12-8-16
Date of Expedited Review

Julius Linn/VMC
Signature of Chair, Vice-Chair or Designee

12-8-16
Date

Informed Consent Document

TITLE OF RESEARCH:	A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers
IRB PROTOCOL NO.:	X161103004
INVESTIGATOR:	Yolanda Harris
SPONSOR:	DAISY (Diseases Attacking the Immune System) Foundation

Purpose of the Research

I am asking you to take part in a research study. The purpose of this research is to explore caregiver perspectives of how family factors influence adaptation in families of children with pediatric onset multiple sclerosis (POMS) seen at a southeastern specialty center. The study will also help to determine the needs of families of children that are newly diagnosed with POMS in order to aid in developing effective coping behaviors that will promote healthy and positive family relationships.

Explanation of Procedures

This study will involve 20 caregivers of children between the ages of 0 and 23 with a diagnosis of pediatric onset multiple sclerosis (POMS). You are being asked to be in the study because you are caregiver of a child with pediatric onset multiple sclerosis and you are at least 18 years of age or older. If you decide to participate in the study, you will be agreeing to take part in a one-on-one interview. The interview can be in person or it can be over the phone. The choice is up to you. Before the interview, you will be asked to complete a brief questionnaire as well as a sociodemographic form giving a description of you, your family and the child with POMS. Your name or identifying information will not be included on the questionnaire or the demographic form. The interview will be conducted in a private room or office and will take approximately 1 hour. The socio-demographic form will take approximately 15 to 20 minutes to complete and the family hardiness index will take approximately 10 minutes to complete.

The one-on-one interview will be audio recorded for research purposes only. All recordings will be stored in a password protected file on a computer in the investigator's office and destroyed once they have been transcribed. Only members of the research team will have access to the tape recording and transcript of the interview.

Risks and Discomforts

Risk to you for being in the study is minimal. You may experience some emotional discomfort as you talk about certain situations. However, you may feel better by having the opportunity to talk about your experiences. If you experience any discomfort or distress during the interview, a follow-up phone call will be made within twenty-four hours of the interview by the investigator.

A referral to counseling will be made if the investigator feels that you are still in distress when the phone call is made.

There is a potential for loss of confidentiality. To protect your confidentiality, your name will not be mentioned during the interview or written on any questionnaires.

Benefits

Although your participation in this study may not have a direct benefit, it may provide you with an opportunity to talk about you and your family's experiences and provide emotional release which may prove therapeutic. Your participation in this study may also benefit other families who have a child with POMS, as the results of this will be used to develop future programs for families of children with POMS that will encourage positive adaptive behaviors, promote effective family communication and foster strengthened relationships among families.

Alternatives

This study involves you participating in a one-on-one interview (in person or over the phone) with the investigator. You have the alternative to not participate in the study.

Confidentiality

Information obtained about you for this study will be kept confidential to the extent allowed by law. However, research information that identifies you may be shared with the UAB Institutional Review Board (IRB) and others who are responsible for ensuring compliance with laws and regulations related to research, including people on behalf of the UAB School of Nursing, the DAISY Foundation, and the Office for Human Research Protections (OHRP). The information from the research may be published for scientific purposes; however, your identity will not be given out. The names of participants will **NOT** be included in any publications or presentations. Study results will be reported in a way that makes it impossible to identify individual people.

If any part of study takes place at Children's of Alabama this consent document will be placed in your file at that facility. The document will become part of your child's medical record chart.

The researcher will refer those participants who remain in distress at follow-up to social services for assistance in seeking counseling. The referral will remain confidential.

By law, any instance of reported physical or sexual abuse involving minors is required to be reported to the appropriate authorities. Only members of the research team will know your names or contact you. No names will be attached to any data records; these will be coded in a way that makes it impossible to identify individual participants. The recorded interview will be typed word for word, omitting any names or other identifying information. All recorded interviews and typed transcripts of the interviews will be stored in a password-protected computer file on the computer in Ms. Harris' office. At the completion of the study, sociodemographic forms will be destroyed. This consent form will be filed in a locked file cabinet in Ms. Harris' office.

Voluntary Participation and Withdrawal

Taking part in this study is your choice. There will be no penalty if you decide not to be in the study. You are free to withdraw from this research study at any time. Your choice not to participate or to withdraw from the study will not affect any services you are now receiving.

Cost of Participation

There will be no cost to you for taking part in this study.

Payment for Participation in Research

You will be paid $40 cash for participation in the study at the end of the individual interview and one both the socio-demographic and Family Hardiness Index® are complete. This money is to reimburse you for your time and the contribution you made to the study.

Questions

If you have any questions, concerns, or complaints about the research, you may contact Ms. Yolanda Harris. She will be glad to answer any of your questions. Ms. Harris' number is 205-613-5872.

If you have questions about your rights as a research participant, or concerns or complaints about the research, you may contact the UAB Office of the IRB (OIRB) at (205) 934-3789 or toll free at 1-855-860-3789. Regular hours for the OIRB are 8:00 a.m. to 5:00 p.m. CT, Monday through Friday. You may also call this number in the event the research staff cannot be reached or you wish to talk to someone else.

Legal Rights

You are not waiving any of your legal rights by signing this informed consent document.

Signatures

Your signature below indicates you that you have read (or been read) the information provided above and agree to participate in this study. You will receive a copy of this signed consent form.

Signature of Participant Date

Signature of Investigator or Person Obtaining Consent Date

University of Alabama at Birmingham
AUTHORIZATION FOR USE/DISCLOSURE OF
PROTECTED HEALTH INFORMATION (PHI) FOR RESEARCH

Participant Name: _____
Research Protocol: A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers
UAB IRB Protocol Number: X161103004
Principal Investigator: Yolanda Harris
Sponsor: Daisy Foundation

What is the purpose of this form? You are being asked to sign this form so that UAB may use and release your protected health information for research. Participation in research is voluntary. If you choose to participate in the research, you must sign this form so that your protected health information may be used for the research.

Why do the researchers want my protected health information? The researchers want to use your protected health information as part of the research protocol listed above and as described to you in the informed consent.

What protected health information do the researchers want to use? All medical information, including but not limited to information and/or records of any diagnosis or treatment of disease or condition, which may include sexually transmitted diseases (e.g., HIV, etc.) or communicable diseases, drug/alcohol dependency, etc.; all personal identifiers, including but not limited to your name, social security number, medical record number, date of birth, dates of service, etc.; any past, present, and future history, examinations, laboratory results, imaging studies and reports and treatments of whatever kind, including but not limited to drug/alcohol treatment, psychiatric/psychological treatment; financial/billing information, including but not limited to copies of your medical bills, and any other information related to or collected for use in the research protocol, regardless of whether the information was collected for research or non-research (e.g., treatment) purposes.

Who will disclose, use and/or receive my protected health information? All individuals/entities listed in the informed consent documents, including but not limited to, the physicians, nurses and staff and others performing services related to the research (whether at UAB or elsewhere); other operating units of UAB, HSF, UAB Highlands, Children's of Alabama, Eye Foundation Hospital, and the Jefferson County Department of Health, as necessary for their operations; the IRB and its staff; the sponsor of the research and its employees and agents, including any CRO; and any outside regulatory agencies, such as the Food and Drug Administration, providing oversight or performing other legal and/or regulatory functions for which access to participant information is required.

How will my protected health information be protected once it is given to others? Your protected health information that is given to the study sponsor will remain private to the extent possible, even though the study sponsor is not required to follow the federal privacy laws. However, once your information is given to other organizations that are not required to follow federal privacy laws, we cannot assure that the information will remain protected.

How long will this Authorization last? Your authorization for the uses and disclosures described in this Authorization does not have an expiration date.

Can I cancel this Authorization? You may cancel this Authorization at any time by notifying the Principal Investigator, in writing, referencing the research protocol and IRB Protocol Number. If you cancel this Authorization, the study doctor and staff will not use any new health information for research. However, researchers may continue to use the protected health information that was provided before you cancelled your authorization.

Can I see my protected health information? You have a right to request to see your protected health information. However, to ensure the scientific integrity of the research, you will not be able to review the research information until after the research protocol has been completed.

Signature of participant: _____ Date: _____

or participant's legally authorized representative: _____ Date: _____

Printed Name of participant's representative: _____

Relationship to the participant: _____

APPENDIX M

SOCIO-DEMOGRAPHIC FORM

ID# _____

SOCIODEMOGRAPHIC FORM

ABOUT YOU

1. A. Age _____

2. What is your relationship to the child that has pediatric onset multiple sclerosis (POMS)?

   □ A. Mother □ C. Step-mother □ E. Guardian
   □ B. Father □ D. Step-father □ F. Other _____

3. What is your highest level of education that you have completed?

   □ A. Grammar school (8th grade or less)
   □ B. Some high school (grade 9-11)
   □ C. High School Diploma or GED
   □ D. Some College, no degree
   □ E. Associates Degree (2 year college)
   □ F. Bachelor's Degree (4 years of College)
   □ G. Technical/trade/vocational training
   □ H. Master's Degree
   □ I. Doctorate Degree
   □ J. Professional Degree (MD, DO, JD, Etc.)

4. What is your marital status?

   □ A. Married/Domestic partnership
   □ B. Single
   □ C. Widowed
   □ D. Separated
   □ E. Divorced
   □ F. Living with partner but not married

5. A. How long have you been in your current marital status? (# in years) _____

6. What is your current employment status?

   □ A. Employed for wages # Hours/ week _____
   □ B. Self-employed # Hours/week _____
   □ C. Student #Hours/week _____
   □ D. Out of work
   □ E. Homemaker
   □ F. Military
   □ G. Retired
   □ H. Unable to work

7. How long have you been in your current employment status? A. _____# months B. _____# years

8. Are you under the care of a healthcare provider for any health care issues?

   □ A. Yes If yes, for what issues? _____
   □ B. No

9. What is your current annual household income (before taxes)?

   □ A. Under $10,000 □ B. $10,000-19,000 □ C. $20,000-29,000

ID# _____

□ D. $30,000-39,000
□ E. $40,000-49,000
□ F. $50,000-74,000
□ G. $75,000-99,000
□ H. $100,000-150,000
□ I. Over $150,000

10. What is your race?

□ A. White
□ B. Black/African American
□ C. American Indian/Native American
□ D. Alaskan Native
□ E. Unknown
□ F. Asian
□ G. Native Hawaiian/Pacific Islander
□ H. Hispanic/Latino
□ I. Multi-racial

11. What is your family's religious preference?

□ A. Baptist
□ B. African Methodist Episcopal (A.M.E)
□ C. Christian Methodist Episcopal (C.M.E.)
□ D. Jehovah Witness
□ E. Pentecostal/Holiness
□ F. Catholic
□ G. Lutheran
□ H. Non-denominational
□ I. Seventh Day Adventist
□ J. Islam
□ K. Other _____
□ L. Non-affiliated

FAMILY STRUCTURE

ABOUT THE OTHER ADULTS IN THE HOUSEHOLD

Who are the adults living in the household?

12. □ A. Adult #1 Age _____

13. Relationship to You

□ A. Spouse □ B. Sibling □ C. Parent □ D. In-Law □ E. Other _____

14. Is this adult under the care of a health professional for any medical condition?

□ A. Yes For what condition(s)? _____
□ B. No

15. What is the current occupation of this adult?

□ A. Employed for wages Hours per week (7 days) _____
□ B. Self-employed Hours per week (7 days) _____
□ C. Student Hours per week (7 days) _____
□ D. Out of work
□ E. Homemaker
□ F. Military
□ G. Retired
□ H. Unable to work

ID# _____

16. □ A. Adult #2 Age _____

17. Relationship to You

□ A. Spouse □ B. Sibling □ C. Parent □ D. In-Law □ E. Other _____

18. Is this person under the care of a health professional for any medical condition?

□ A. Yes For what condition(s)? _____
□ B. No

19. What is the current occupation of this person?
□ A. Employed for wages Hours per week (7 days) _____
□ B. Self-employed Hours per week (7 days) _____
□ C. Student Hours per week (7 days) _____
□ D. Out of work
□ E. Homemaker
□ F. Military
□ G. Retired
□ H. Unable to work

20. □ A. Adult #3 Age _____

21. Relationship to You

□ A. Spouse □ B. Sibling □ C. Parent □ D. In-Law □ E. Other _____

22. Is this person under the care of a health professional for any medical condition?

□ A. Yes For what condition(s)? _____
□ B. No

23. What is the current occupation of this person?
□ A. Employed for wages Hours per week (7 days) _____
□ B. Self-employed Hours per week (7 days) _____
□ C. Student Hours per week (7 days) _____
□ D. Out of work
□ E. Homemaker
□ F. Military
□ G. Retired
□ H. Unable to work

ABOUT THE CHILD WITH POMS

24. A. What is the child with a POMS current age? _____

ID# _____

25. What is the child with POMS sex? □ A. Female □ B. Male

26. What is the child's race?

□ A. White
□ B. Black/African American
□ C. American Indian/Native American
□ D. Unknown
□ E. Alaskan Native
□ F. Asian
□ G. Native Hawaiian/Pacific Islander
□ H. Hispanic/Latino
□ I. Multi-racial

27. A. Age at time of Diagnosis ____

28. A. _____# of Months B. _____ # of Years child was experiencing symptoms prior to diagnosis

29. How often is your child seen at the CPODD for routine care?

□ A. Monthly □ B. Every three months □ C. Every six months □ D. Annually

30. How many miles do you travel from you home to CPODD?

□ A. <50 □ B. 51-70 □ C. 71-100 □ D. 100-200 □ E. >200

31. Is the child's primary MS care provided at CPODD? □ A. Yes □ B. No

If no, the health professional that provides primary care is one of the following:

□ A. Pediatric neurologist
□ B. Adult Neurologist
□ C. Nurse Practitioner
□ D. Family Medicine Doctor
□ E. Pediatrician
□ F. Other _____

32. How many miles do you travel from you home to the provider that provides primary MS care?

□ A. <50 □ B. 51-70 □ C. 71-100 □ D. 100-200 □ E. >200

33. Which medication does your child take for the treatment of MS progression?

□ A. Copaxone daily
□ B. Copaxone 3/week
□ C. Betaseron
□ D. Glatopa
□ E. Extavia
□ F. Rebif
□ G. Avonex
□ H. Zinbryta
□ I. Tysabri
□ J. Tecfidera
□ K. Gilenya
□ L. Plegridy

34. Is the child currently on medications for symptom management? □ A. Yes □ B. No

35. If yes, check all that apply:

□ A. Bladder dysfunction
□ B. Bowel management
□ C. Fatigue
□ D. Tremor
□ E. Spasticity
□ F. Nerve pain
□ G. Dizziness
□ H. Memory impairment
□ I. Attention/concentration
□ J. Depression
□ K. Anxiety

36. Within the first 2 years of diagnosis, how many flare-ups did the child experience that required the following?

ID# _____

A. ____# requiring doctor's outpatient infusion
B. ____ # requiring outpatient services (PT, OT, speech, pool therapy, etc.)
C. ____ # requiring an overnight stay at the hospital
D. ____ # requiring 2-5 day hospital stay
E. ____ # requiring 1 to 2 week hospital stay
F. ____ # requiring more than 2 weeks but less than one month
G. ____ # requiring an admission to Rehabilitation

37. A. What is the total number of flare-ups that child has experienced since the diagnosis that required a doctor's visit, professional treatment, care coordination or hospitalization? _____

38. Has the child experience any permanent symptoms as a result of MS?

□ A. Blindness/severe visual dysfunction
□ B. Loss of bladder/bowel control
□ C. Need for wheelchair/walker
□ D. Severe cognitive dysfunction
□ E. Deafness
□ F. Impaired swallowing
□ G. Impaired use of upper extremities
□ H. Heat/cold sensitivity
□ I. Mood alterations
□ J. Depression
□ K. Anxiety
□ L. Impaired attention/concentration

About the Other Children in the Household

39. □ A. Child # 1 Age _____

40. Sex: □ A. Male □ B. Female

41. What is your relationship with the child?
□ A. Parent
□ B. Step-parent
□ C. Grandparent
□ D. Guardian
□ E. Aunt/uncle
□ F. Other_____

42. Is this child under the care of a health professional for a medical condition?

□ A. Yes If yes, for what condition? _____

□ B. No

43. □ A. Child # 2 Age _____

44. Sex: □ A. Male □ B. Female

45. What is your relationship with the child?
□ A. Parent
□ B. Step-parent
□ C. Grandparent
□ D. Guardian
□ E. Aunt/uncle
□ F. Other_____

ID# _____

46. Is this child under the care of a health professional for a medical condition?

□ A. Yes If yes, for what condition? _____
□ B. No

47. □ A. Child # 3 Age _____

48. Sex: □ A. Male □ B. Female

49. What is your relationship with the child?

□ A. Parent
□ B. Step-parent
□ C. Grandparent
□ D. Guardian
□ E. Aunt/uncle
□ F. Other_____

50. Is this child under the care of a health professional for a medical condition?

□ A. Yes If yes, for what condition? _____

□ B. No

APPENDIX N

INTERVIEW SCRIPT

Interview Script for the ADAPT2POMS Study

Interviewer: Thank you again for agreeing to participate in the one-on-one interview about family factors and how they influence the family's adaptation to pediatric onset multiple sclerosis. Before we start the interview, I have requests of you first. I am asking you to please speak clearly into the recorder that your interview can be captured. Please share your personal views and experiences don't worry about my thoughts and opinions. There's no right or wrong answer. Your ideas and experience are very important to us and will help us to understand the experiences of families with child with multiple sclerosis. Anything you tell me will not be shared with anyone else outside of the research team. Everything is confidential and your interview will be recorded. I can't share anything that you tell me with your other health care providers or your child because I signed a confidentiality statement to ensure that you are protected. Please feel free to share everything and anything that you think is relevant to what we are discussing in the interview because what you have to say is very important to us for this study.

Interviewer: Now let's begin with a few questions about you and your family.
Tell me about yourself.
Probes:

- What do you like to do for fun?
- What hobbies do you have?
- Tell me what you and your family like to do as a group.
- Do you eat dinner together?
- Do you have any family rituals?
- Do you attend religious services together?
- Do you take trips to see family or go on vacation together?

Interviewer: Tell me how you deal with stress.

Probe:

- What do you do to relieve stress?

Interviewer: Now let's move on to more specific topics about your family experience with a child with pediatric multiple sclerosis.
Tell me about the family's experience with receiving the diagnosis.

Probes:

- Describe your you and your family's emotions when you first heard the words "multiple sclerosis"
- Explain what immediate changes took place
- Explain what changes were gradual.
- Explain what changes were unexpected

Interviewer: Tell me about any stress or strain that your family may have been dealing with prior to the diagnosis being made.

Probes:

- Describe any financial issues you were dealing with prior to the diagnosis such as change in job status, demands on the family.
- Describe any health issues that your family dealt with prior to the diagnosis.
- Describe the relationships with family members prior to the diagnosis.

Interviewer: Tell me about any changes that the family has had to face since the diagnosis.

Probes:

- Describe the changes in the role of family members after the diagnosis.
- Describe the amount of time you spend per day or week providing MS specific care to the child with POMS.
- Describe how the diagnosis has affected your family's relationships within the home.
- Describe how the diagnosis has affected the family's relationships with others within the home.
- Describe your family's biggest challenge that had to be overcome after the diagnosis.
- Describe how the family is affected during a relapse.
- Describe how the family is affected after a relapse and the child need to become stable due to follow-up, rehab, etc.

Interviewer: Tell me about your personal strengths.

Probes:

- Describe your current level of understanding of what an MS diagnosis means for your child and your family.
- Explain how this understanding has changed over time.
- Describe your child's current level of understanding of what an MS diagnosis means for them.
- Explain how this understanding has changed over time.

Interviewer: Tell me about the resources that you have that help you with raising a child with POMS.

Probes:

- Describe the medical resources that you have to help you deal with a diagnosis.
- Describe the community resources that your family can call upon or have access to help deal with a diagnosis.
- Describe any personal resources that you have to help deal with a diagnosis.
- Describe your access to technology that aids in your access to information and support.

Interviewer: Tell me how your family view your experiences of raising a child with POMS.

Probes:

- Tell me what your family thinks about raising a child with POMS.

Interviewer: Tell me about how your family manages a child with POMS.
Probes:

- Describe how your family manages your day to day life.
- Describe how your family manages when a crisis or unexpected event happens.

Interviewer: Tell me how your family copes with raising a child with POMS.

Probes:

- Describe how your family communicates with each other on a daily basis.
- Describe how communication within your family changes during an unexpected flare-up or unexpected medical event related to your child's illness.

Interviewer: Tell me how you achieve a balance between the needs of the child, the family and other family members.

Probes:

- If you had to describe the top strategies that you have developed or used as a family to manage the demands of this illness and the needs of the family, what would they be?

Interviewer: Now. Let's sum up your experiences.
Probes:

- If you had to write a book about your family's experience on raising a child with POMS, what would the title of that book be?
- What would be the main message of your book?
- What advice do you have for other families who have children recently diagnosed with POMS?
- What advice do you have to healthcare providers who care for children with POMS and their families?

Interviewer: Thank you for sharing your family's experiences. Is there anything else that you think I should know that we didn't talk about before we end the session?

APPENDIX O

INFORMED CONSENT AND AUTHORIZATION

Informed Consent Document

TITLE OF RESEARCH:	A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers
IRB PROTOCOL NO.:	X161103004
INVESTIGATOR:	Yolanda Harris
SPONSOR:	UAB School of Nursing

Purpose of the Research

I am asking you to take part in a research study. The purpose of this research is to explore caregiver perspectives of how family factors influence adaptation in families of children with pediatric onset multiple sclerosis (POMS) seen at a southeastern specialty center. The study will also help to determine the needs of families of children that are newly diagnosed with POMS in order to aid in developing effective coping behaviors that will promote healthy and positive family relationships.

Explanation of Procedures

This study will involve 20 family caregivers who live in the household with children between the ages of 0 and 23 with a diagnosis of pediatric onset multiple sclerosis (POMS). You are being asked to be in the study because you are family caregiver of a child with pediatric onset multiple sclerosis and you are at least 18 years of age or older. If you decide to participate in the study, you will be agreeing to take part in a one-on-one interview. The interview can be in person or it can be over the phone. The choice is up to you. Before the interview, you will be asked to complete a sociodemographic form giving a description of you, your family and the child with POMS. Your name or identifying information will not be included on the demographic form. The interview will be conducted in a private room or office and will take approximately 1 hour. The socio-demographic form will take approximately 15 to 20 minutes to complete.

The one-on-one interview will be audio recorded for research purposes only. All recordings will be stored in a password protected file on a computer in the investigator's office and destroyed once they have been transcribed. Only members of the research team will have access to the tape recording and transcript of the interview.

Risks and Discomforts

Risk to you for being in the study is minimal. You may experience some emotional discomfort as you talk about certain situations. However, you may feel better by having the opportunity to talk about your experiences. If you experience any discomfort or distress during the interview, a follow-up phone call will be made within twenty-four

hours of the interview by the investigator. A referral to counseling will be made if the investigator feels that you are still in distress when the phone call is made.

There is a potential for loss of confidentiality. To protect your confidentiality, your name will not be mentioned during the interview or written on the socio-demographic form.

Benefits

Although your participation in this study may not have a direct benefit, it may provide you with an opportunity to talk about you and your family's experiences and provide emotional release which may prove therapeutic. Your participation in this study may also benefit other families who have a child with POMS, as the results of this will be used to develop future programs for families of children with POMS that will encourage positive adaptive behaviors, promote effective family communication and foster strengthened relationships among families.

Alternatives

This study involves you participating in a one-on-one interview (in person or over the phone) with the investigator. You have the alternative to not participate in the study.

Confidentiality

Information obtained about you for this study will be kept confidential to the extent allowed by law. However, research information that identifies you may be shared with the UAB Institutional Review Board (IRB) and others who are responsible for ensuring compliance with laws and regulations related to research, including people on behalf of the UAB School of Nursing, and the Office for Human Research Protections (OHRP). The information from the research may be published for scientific purposes; however, your identity will not be given out. The names of participants will **NOT** be included in any publications or presentations. Study results will be reported in a way that makes it impossible to identify individual people.

If any part of study takes place at Children's of Alabama this consent document will be placed in your file at that facility. The document will become part of your child's medical record chart.

The researcher will refer those participants who remain in distress at follow-up to social services for assistance in seeking counseling. The referral will remain confidential.

By law, any instance of reported physical or sexual abuse involving minors is required to be reported to the appropriate authorities. Only members of the research team will know your names or contact you. No names will be attached to any data records; these will be coded in a way that makes it impossible to identify individual participants. The recorded interview will be typed word for word, omitting any names or other identifying

information. All recorded interviews and typed transcripts of the interviews will be stored in a password-protected computer file on the computer in Ms. Harris' office. At the completion of the study, sociodemographic forms will be destroyed. This consent form will be filed in a locked file cabinet in Ms. Harris' office.

Voluntary Participation and Withdrawal

Taking part in this study is your choice. There will be no penalty if you decide not to be in the study. You are free to withdraw from this research study at any time. Your choice not to participate or to withdraw from the study will not affect any services you are now receiving.

Cost of Participation

There will be no cost to you for taking part in this study.

Payment for Participation in Research

You will be paid $25gift card for participation in the study at the end of the individual interview and after the socio-demographic are complete. This money is to reimburse you for your time and the contribution you made to the study.

Questions

If you have any questions, concerns, or complaints about the research, you may contact Ms. Yolanda Harris. She will be glad to answer any of your questions. Ms. Harris' number is 205-613-5872.

If you have questions about your rights as a research participant, or concerns or complaints about the research, you may contact the UAB Office of the IRB (OIRB) at (205) 934-3789 or toll free at 1-855-860-3789. Regular hours for the OIRB are 8:00 a.m. to 5:00 p.m. CT, Monday through Friday. You may also call this number in the event the research staff cannot be reached or you wish to talk to someone else.

Legal Rights

You are not waiving any of your legal rights by signing this informed consent document.

Signatures

Your signature below indicates you that you have read (or been read) the information provided above and agree to participate in this study. You will receive a copy of this signed consent form.

Signature of Participant	Date
Signature of Investigator or Person Obtaining Consent	Date

University of Alabama at Birmingham
AUTHORIZATION FOR USE/DISCLOSURE OF
PROTECTED HEALTH INFORMATION (PHI) FOR RESEARCH

Participant Name: _____
Research Protocol: A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers

UAB IRB Protocol Number: X161103004
Principal Investigator: Yolanda Harris
Sponsor: UAB School of Nursing

What is the purpose of this form? You are being asked to sign this form so that UAB may use and release your protected health information for research. Participation in research is voluntary. If you choose to participate in the research, you must sign this form so that your protected health information may be used for the research.

Why do the researchers want my protected health information? The researchers want to use your protected health information as part of the research protocol listed above and as described to you in the informed consent.

What protected health information do the researchers want to use? All medical information, including but not limited to information and/or records of any diagnosis or treatment of disease or condition, which may include sexually transmitted diseases (e.g., HIV, etc.) or communicable diseases, drug/alcohol dependency, etc.; all personal identifiers, including but not limited to your name, social security number, medical record number, date of birth, dates of service, etc.; any past, present, and future history, examinations, laboratory results, imaging studies and reports and treatments of whatever kind, including but not limited to drug/alcohol treatment, psychiatric/psychological treatment; financial/billing information, including but not limited to copies of your medical bills, and any other information related to or collected for use in the research protocol, regardless of whether the information was collected for research or non-research (e.g., treatment) purposes.

Who will disclose, use and/or receive my protected health information? All Individuals/entities listed in the informed consent documents, including but not limited to, the physicians, nurses and staff and others performing services related to the research (whether at UAB or elsewhere); other operating units of UAB, HSF, UAB Highlands, Children's of Alabama, Eye Foundation Hospital, and the Jefferson County Department of Health, as necessary for their operations; the IRB and its staff; the sponsor of the research and its employees and agents, including any CRO; and any outside regulatory agencies, such as the Food and Drug Administration, providing oversight or performing other legal and/or regulatory functions for which access to participant information is required.

How will my protected health information be protected once it is given to others? Your protected health information that is given to the study sponsor will remain private to the extent possible, even though the study sponsor is not required to follow the federal privacy laws. However, once your information is given to other organizations that are not required to follow federal privacy laws, we cannot assure that the information will remain protected.

How long will this Authorization last? Your authorization for the uses and disclosures described in this Authorization does not have an expiration date.

Can I cancel this Authorization? You may cancel this Authorization at any time by notifying the Principal

Investigator, in writing, referencing the research protocol and IRB Protocol Number. If you cancel this Authorization, the study doctor and staff will not use any new health information for research. However, researchers may continue to use the protected health information that was provided before you cancelled your authorization.

Can I see my protected health information? You have a right to request to see your protected health information. However, to ensure the scientific integrity of the research, you will not be able to review the research information until after the research protocol has been completed.

Signature of participant: _____ Date: _____

or participant's legally authorized representative: _____ Date: _____

Printed Name of participant's representative: _____

Relationship to the participant: _____

APPENDIX P

VERBAL CONSENT AND PHONE INTERVIEW SCRIPT

Phone Script for Verbal Consent and Telephone Interview

PI: Hello. This is Yolanda Harris. As you know, I am conducting interviews with the caregivers of children with pediatric onset multiple sclerosis to identify factors that influence how a family adapts to the changes that come with this diagnosis. I am conducting this interview as part of research for my PhD studies at UAB School of Nursing in Birmingham, AL.

PI: *I have invited you and other caregivers of children to do a one-on-one interview with me that will take about 60 minutes. In the interview, I will ask you questions about you, your family, your child with MS and the other family members that live in your household. I will talk to you about things such as your relationships with family, friends, and spouse/significant other; changes that have occurred in your family, personal strengths, resources and experiences with having a child with MS. The interview will be audiotaped so that I can record your responses. I may also take some notes as we talk. The recording will be for research purposes only and destroyed after the research is over. Do you have any questions at this point?*

Pause to allow the participant to respond. The PI will answer any study-related questions posed by the participant.

PI: *It is likely that there will be minimal or no discomfort associated with your participation in this study. You may feel emotional with some questions that are asked. In that instance I will pause and let you regroup your thought and emotions. However, you may feel better by having the opportunity to talk about your experience. You do not need to answer questions that you do not want to answer or that make you feel uncomfortable…. And you can withdraw (stop taking part) from the study at any time. Do you have any questions?*

Pause to allow the participant to respond. The PI will answer any study-related questions posed by the participant.

PI: *Although you may not have a direct benefit from participating in this study, the information that you share will inform the development of programs to assist other families of children with POMS.*

PI: *There will be no cost to you for taking part in this study. Once the questionnaires and interview are completed, you will be paid $25 cash for your participation in the study. This money is to reimburse you for your time and the contribution you made to the study.*

PI*:* *I will maintain your confidentiality by storing any document with your name on it, such as the consent form, in a locked file cabinet in my office at the UAB School of Nursing. To maintain your confidentiality during the interview, I will not use your real name. Any data from this research study that is shared or published will be the combined data of all participants. That means it will be reported for the whole group not for individual persons. Do you have any questions about this?*

Pause to allow the participant to respond. The PI will answer any study-related questions posed by the participant.

PI: *Your participation in this study is voluntary. You can decide to stop at any time, even part-way through the interview for whatever reason. Additionally, it is up to you whether you answer a question or not. If you decide not to answer a question or choose to stop participating in the study, there will be no consequences to you. Your choice to stop participating in the study will not affect any of the services you are receiving from the clinic. If you decide to stop, we will ask you how you would like us to handle the data collected up to that point. This could include returning it to you, destroying it or using it in the final analysis of the data.*

PI*:* *This study has been reviewed and cleared by the UAB IRB. If you have concerns or questions about your rights as a participant or about the way the study is conducted, you may contact:*

UAB office of the IRB
AB 470
1720 2nd Avenue South
Birmingham, AL 35294-0104
Phone: 205-934-3789
Hours: 8:00 am -5:00 pm CST, Monday thru Friday

PI*:* *Do you have any questions or would like any additional details*? *If you have any other questions about this study or would like more information you can call me at* ***(205)613-5872.***

PI waits for questions and answers

PI: *Do you agree to participate in this study knowing that you can withdraw at any point with no consequences to you?*

[If yes, the PI begins the interview.]

[If no, the PI thanks the participant for his/her time.]

Interviewer: *Thank you again for agreeing to participate in the one-on-one interview about family factors and how they influence the family's adaptation to pediatric onset multiple sclerosis. Before we start the interview, I have requests of you first. I am asking you to please speak clearly into the recorder that your interview can be captured. Please share your personal views and experiences don't worry about my thoughts and opinions. There's no right or wrong answer. Your ideas and experience are very important to us and will help us to understand the experiences of families with child with multiple sclerosis. Anything you tell me will not be shared with anyone else outside of the research team. Everything is confidential and your interview will be recorded. I can't share anything that you tell me with your other health care providers or your child because I signed a confidentiality statement to ensure that you are protected. Please feel free to share everything and anything that you think is relevant to what we are discussing in the interview because what you have to say is very important to us for this study.*

Interviewer: *Now let's begin with a few questions about you and your family. Tell me about yourself.*
Probes:

- *What do you like to do for fun?*
- *What hobbies do you have?*
- *Tell me what you and your family like to do as a group.*
- *Do you eat dinner together?*
- *Do you have any family rituals?*
- *Do you attend religious services together?*
- *Do you take trips to see family or go on vacation together?*

Interviewer: *Tell me how you deal with stress.*

Probe:

- *What do you do to relieve stress?*

Interviewer: *Now let's move on to more specific topics about your families. experience with a child with pediatric multiple sclerosis.*

Tell me about the family's experience with receiving the diagnosis.

Probes:

- *Describe your you and your family's emotions when you first heard the words "multiple sclerosis"*
- *Explain what immediate changes took place*
- *Explain what changes were gradual.*
- *Explain what changes were unexpected*

Interviewer: *Tell me about any stress or strain that your family may have been dealing with prior to the diagnosis being made.*

Probes:

- *Describe any financial issues you were dealing with prior to the diagnosis such as change in job status, demands on the family.*
- *Describe any health issues that your family dealt with prior to the diagnosis.*
- *Describe the relationships with family members prior to the diagnosis.*

Interviewer: *Tell me about any changes that the family has had to face since the diagnosis.*

Probes:

- *Describe the changes in the role of family members after the diagnosis.*
- *Describe the amount of time you spend per day or week providing MS specific care to the child with POMS.*
- *Describe how the diagnosis has affected your family's relationships within the home.*
- *Describe how the diagnosis has affected the family's relationships with others within the home.*
- *Describe your family's biggest challenge that had to be overcome after the diagnosis.*
- *Describe how the family is affected during a relapse.*
- *Describe how the family is affected after a relapse and the child need to become stable due to follow-up, rehab, etc.*

Interviewer: *Tell me about your personal strengths.*

Probes:

- *Describe your current level of understanding of what an MS diagnosis means for your child and your family.*
- *Explain how this understanding has changed over time.*
- *Describe your child's current level of understanding of what an MS diagnosis means for them.*
- *Explain how this understanding has changed over time.*

Interviewer: *Tell me about the resources that you have that help you with raising a child with POMS.*

Probes:

- *Describe the medical resources that you have to help you deal with a diagnosis.*
- *Describe the community resources that your family can call upon or have access to help deal with a diagnosis.*
- *Describe any personal resources that you have to help deal with a diagnosis.*
- *Describe your access to technology that aids in your access to information and support.*

Interviewer: *Tell me how your family view your experiences of raising a child with POMS.*

Probes:

- *Tell me what your family thinks about raising a child with POMS.*
- *What does your family feel about the future of the child with POMS?*

Interviewer: *Tell me about how your family manages a child with POMS.*
Probes:

- *Describe how your family manages your day to day life.*
- *Describe how your family manages when a crisis or unexpected event happens.*

Interviewer: *Tell me how your family copes with raising a child with POMS.*

Probes:

- *Describe how your family communicates with each other on a daily basis.*
- *Describe how communication within your family changes during an unexpected flare-up or unexpected medical event related to your child's illness.*

Interviewer: *Tell me how you achieve a balance between the needs of the child, the family and other family members.*

Probes:

- *If you had to describe the top strategies that you have developed or used as a family to manage the demands of this illness and the needs of the family, what would they be?*

Interviewer: *Now. Let's sum up your experiences.*
Probes:

- *If you had to write a book about your family's experience on raising a child with POMS, what would the title of that book be?*
- *What would be the main message of your book?*
- *What advice do you have for other families who have children recently diagnosed with POMS?*
- *What advice do you have to healthcare providers who care for children with POMS and their families?*

Interviewer: *Thank you for sharing your family's experiences. Is there anything else that you think I should know that we didn't talk about before we end the session?*

APPENDIX Q

APPOINTMENT REMINDER SCRIPT

Appointment Reminder Script

*This script will be used as appointment reminders for participants to be called one week prior to their scheduled appointment.

PI or research staff: *"Hello name of the participant, this is name of the caller, from the ADAPT2POMS Research Study. I am calling to remind you of your upcoming appointment for your one-on-one interview that is scheduled for day of the week, month, day, at time."*
Do you have any questions or concerns regarding your appointment?

Wait for a response from the potential participant.
If the participant has questions, answer the questions, and then take them for their time.
If the participant does not have any questions, thank them for their time.

APPENDIX R

AMENDMENT TO IRB APPROVAL

UAB

Project Revision/Amendment Form

irb

Form version: June 26, 2012

In MS Word, click in the white boxes and type your text; double-click checkboxes to check/uncheck.

- Federal regulations require IRB approval before implementing proposed changes. See Section 14 of the IRB Guidebook for Investigators for additional information.
- Change means any change, in content or form, to the protocol, consent form, or any supportive materials (such as the Investigator's Brochure, questionnaires, surveys, advertisements, etc.). See Item 4 for more examples.

1. Today's Date	02/01/2017	30860

2. Principal Investigator (PI)

Name (with degree)	Yolanda Harris, MSN, CPNP-AC/Nursing PhD Student	Blazer ID	yoharris
Department	School of Nursing	Division (if applicable)	
Office Address	1720 2nd Ave S, NB 418	Office Phone	205-934-0639
E-mail	yoharris@uab.edu	Fax Number	205-996-7183

Contact person who should receive copies of IRB correspondence (Optional)

Name	Yolanda Harris	E-Mail	
Phone	205-613-5872	Fax Number	
Office Address (if different from PI)			

3. UAB IRB Protocol Identification

3.a. Protocol Number	X161103004
3.b. Protocol Title	A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers

3.c. Current Status of Protocol—Check ONE box at left; provide numbers and dates where applicable

- ☒ **Study has not yet begun** — No participants, data, or specimens have been entered.
- ☐ **In progress, open to accrual** — Number of participants, data, or specimens entered:
- ☐ **Enrollment temporarily suspended by sponsor**
- ☐ **Closed to accrual, but procedures continue as defined in the protocol (therapy, intervention, follow-up visits, etc.)**
  - Date closed:
  - Number of participants receiving interventions:
  - Number of participants in long-term follow-up only:
- ☐ **Closed to accrual, and only data analysis continues**
  - Date closed:
  - Total number of participants entered:

4. Types of Change

Check all types of change that apply, and describe the changes in Item 5.c. or 5.d. as applicable. To help avoid delay in IRB review, please ensure that you provide the required materials and/or information for each type of change checked.

- ☒ **Protocol revision (change in the IRB-approved protocol)**
  In Item 5.c., if applicable, provide sponsor's protocol version number, amendment number, update number, etc.
- ☒ **Protocol amendment (addition to the IRB-approved protocol)**
  In Item 5.c., if applicable, provide funding application document from sponsor, as well as sponsor's protocol version number, amendment number, update number, etc.
- ☒ **Add or remove personnel**
  In Item 5.c., include name, title/degree, department/division, institutional affiliation, and role(s) in research, and address whether new personnel have any conflict of interest. See "Change in Principal Investigator" in the IRB Guidebook if the principal investigator is being changed.
  - ☐ **Add graduate student(s) or postdoctoral fellow(s) working toward thesis, dissertation, or publication**
    In Item 5.c., (a) identify these individuals by name; (b) provide the working title of the thesis, dissertation, or publication; and (c) indicate whether or not the student's analysis differs in any way from the purpose of the research described in the IRB-approved HSP (e.g., a secondary analysis of data obtained under this HSP).
- ☒ **Change in source of funding; change or add funding**
  In Item 5.c., describe the change or addition in detail, include the applicable OSP proposal number(s), and provide a copy of the application as funded (or as submitted to the sponsor if pending). Note that some changes in funding may require a new IRB application.

☐	**Add or remove performance sites** In Item 5.c., identify the site and location, and describe the research-related procedures performed there. If adding site(s), attach notification of permission or IRB approval to perform research there. Also include copy of subcontract, if applicable. If this protocol includes acting as the Coordinating Center for a study, attach IRB approval from any non-UAB site added.
☐	**Add or change a genetic component or storage of samples and/or data component—this could include data submissions for Genome-Wide Association Studies (GWAS)** To assist you in revising or preparing your submission, please see the IRB Guidebook for Investigators or call the IRB office at 934-3789.
☐	**Suspend, re-open, or permanently close protocol to accrual of individuals, data, or samples (IRB approval to remain active)** In Item 5.c., indicate the action, provide applicable dates and reasons for action; attach supporting documentation.
☐	**Report being forwarded to IRB (e.g., DSMB, sponsor or other monitor)** In Item 5.c., include date and source of report, summarize findings, and indicate any recommendations.
☒	**Revise or amend consent, assent form(s)** Complete Item 5.d.
☐	**Addendum (new) consent form** Complete Item 5.d.
☒	**Add or revise recruitment materials** Complete Item 5.d.
☐	**Other (e.g., investigator brochure)** Indicate the type of change in the space below, and provide details in Item 5.c. or 5.d. as applicable. Include a copy of all affected documents, with revisions highlighted as applicable.

▶

5. Description and Rationale

In Item 5.a. and 5.b, check Yes **or** No **and see instructions for** Yes **responses.**
In Item 5.c. and 5.d, describe—and explain the reason for—the change(s) noted in Item 4.

☒Yes ☐No	**5.a. Are any of the participants enrolled as normal, healthy controls?** If yes, describe in detail in Item 5.c. how this change will affect those participants.
☒Yes ☐No	**5.b. Does the change affect subject participation, such as procedures, risks, costs, location of services, etc.?** If yes, FAP-designated units complete a FAP submission and send to fap@uab.edu. Identify the FAP-designated unit in Item 5.c. For more details on the UAB FAP, see www.uab.edu/cto.

5.c. Protocol Changes: In the space below, briefly describe—and explain the reason for—all change(s) to the protocol.

- Personnel was added to help with recruitment and informed consent. Personnel was removed for data analysis as this will now be done by the researcher's PhD mentor, Dr. Childs.
- The funding was changed from The DAISY Foundation to UAB School of Nursing as the grant was not funded by the listed sponsor.
- The purpose and specific aims were modified per the researcher's dissertation committee recommendations.
- The background was modified to include information regarding the impact on families per the researchers' dissertation committee recommendations.
- The Inclusion criteria was modified to specify a "family" caregiver "who lives in the household with a child diagnosed with POMS" as "defined using the 2010 McDonald Criteria" per the researchers' dissertation committee recommendation. This inclusion criteria is more descriptive
- Under the protocol procedures, one of the components of the research study was removed-the Family Hardiness Index. This would make the study mix-methods instead of pure qualitative study. This was removed from this study per the recommendations of the dissertation committee.
- For those participants who chose to complete the interview over the phone, a statement was added to include the use of face-time between the researcher and participant to capture emotions or non-verbal ques that may be otherwise missed if the interview is done by a phone call alone.
- The approximate time was changed to account for the removal of the family hardness index
- Incentive amount was reduced from $40 to $25 since the funding source changed.
- Two additional interview questions were added to the Interview Script to cover material discussed to be important in the researcher's dissertation proposal defense and recommended by the researcher's

dissertation defense committee

- An additional question was added to the Screening Tool to ensure that the inclusion criteria is clear per the dissertation committee recommendations
- The Socio-Demographic Form was updated for formatting and coding purposes only per the recommendation of the dissertation committee

5.d. Consent and Recruitment Changes: In the space below,
(a) describe all changes to IRB-approved forms or recruitment materials and the reasons for them;
(b) describe the reasons for the addition of any materials (e.g., addendum consent, recruitment); and
(c) indicate either how and when you will reconsent enrolled participants or why reconsenting is not necessary (not applicable for recruitment materials).

Also, indicate the number of forms changed or added. For new forms, provide 1 copy. For revised documents, provide 3 copies:
• a copy of the currently approved document (showing the IRB approval stamp, if applicable)
• a revised copy highlighting all proposed changes with "tracked" changes
• a revised copy for the IRB approval stamp.

- The Consent, Authorization and Recruitment Flyer were changed
- On the Consent Form the following was change:
  - Sponsor was changed from DAISY Foundation to UAB School of Nursing; funding was not acquired and study is now self- funded by the researcher
  - "family" is added to the language in the consent –this study will include family caregivers only
  - Any referral to the "Family hardiness Index" was removed since it will no longer be administered
  - The payment amount for the incentive was reduced from $40 to $25 per the decision of the researcher
- On the Authorization Form- the Sponsor was changed to match the consent and HSP
- On the Recruitment Flyer, the following was changed:
  - "family" is added to language in who can participate
  - Removed "clinically definite" from the description of participates. This will be determined during the screening process
  - Removed the "Family Hardiness" information since it will no longer be collected

Signature of Principal Investigator Yolanda Stair Date 02/06/17

FOR IRB USE ONLY

☐ Received & Noted ☑ Approved Expedited* ☐ To Convened IRB

BQuin 2/16/17

Signature (Chair, Vice-Chair, Designee) Date

DOLA 12/8/16

Change to Expedited Category Y / (N) / NA

*No change to IRB's previous determination of approval criteria at 45 CFR 46.111 or 21 CFR 56.111

Informed Consent Document

TITLE OF RESEARCH:	A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers
IRB PROTOCOL NO.:	X161103004
INVESTIGATOR:	Yolanda Harris
SPONSOR:	UAB School of Nursing

Purpose of the Research

I am asking you to take part in a research study. The purpose of this research is to explore caregiver perspectives of how family factors influence adaptation in families of children with pediatric onset multiple sclerosis (POMS) seen at a southeastern specialty center. The study will also help to determine the needs of families of children that are newly diagnosed with POMS in order to aid in developing effective coping behaviors that will promote healthy and positive family relationships.

Explanation of Procedures

This study will involve 20 family caregivers who live in the household with children between the ages of 0 and 23 with a diagnosis of pediatric onset multiple sclerosis (POMS). You are being asked to be in the study because you are family caregiver of a child with pediatric onset multiple sclerosis and you are at least 18 years of age or older. If you decide to participate in the study, you will be agreeing to take part in a one-on-one interview. The interview can be in person or it can be over the phone. The choice is up to you. Before the interview, you will be asked to complete a sociodemographic form giving a description of you, your family and the child with POMS. Your name or identifying information will not be included on the demographic form. The interview will be conducted in a private room or office and will take approximately 1 hour. The socio-demographic form will take approximately 15 to 20 minutes to complete.

The one-on-one interview will be audio recorded for research purposes only. All recordings will be stored in a password protected file on a computer in the investigator's office and destroyed once they have been transcribed. Only members of the research team will have access to the tape recording and transcript of the interview.

Risks and Discomforts

Risk to you for being in the study is minimal. You may experience some emotional discomfort as you talk about certain situations. However, you may feel better by having the opportunity to talk about your experiences. If you experience any discomfort or distress during the interview, a follow-up phone call will be made within twenty-four hours of the interview by the investigator.

A referral to counseling will be made if the investigator feels that you are still in distress when the phone call is made.

There is a potential for loss of confidentiality. To protect your confidentiality, your name will not be mentioned during the interview or written on the socio-demographic form.

Benefits

Although your participation in this study may not have a direct benefit, it may provide you with an opportunity to talk about you and your family's experiences and provide emotional release which may prove therapeutic. Your participation in this study may also benefit other families who have a child with POMS, as the results of this will be used to develop future programs for families of children with POMS that will encourage positive adaptive behaviors, promote effective family communication and foster strengthened relationships among families.

Alternatives

This study involves you participating in a one-on-one interview (in person or over the phone) with the investigator. You have the alternative to not participate in the study.

Confidentiality

Information obtained about you for this study will be kept confidential to the extent allowed by law. However, research information that identifies you may be shared with the UAB Institutional Review Board (IRB) and others who are responsible for ensuring compliance with laws and regulations related to research, including people on behalf of the UAB School of Nursing, , and the Office for Human Research Protections (OHRP). The information from the research may be published for scientific purposes; however, your identity will not be given out. The names of participants will **NOT** be included in any publications or presentations. Study results will be reported in a way that makes it impossible to identify individual people.

If any part of study takes place at Children's of Alabama this consent document will be placed in your file at that facility. The document will become part of your child's medical record chart.

The researcher will refer those participants who remain in distress at follow-up to social services for assistance in seeking counseling. The referral will remain confidential.

By law, any instance of reported physical or sexual abuse involving minors is required to be reported to the appropriate authorities. Only members of the research team will know your names or contact you. No names will be attached to any data records; these will be coded in a way that makes it impossible to identify individual participants. The recorded interview will be typed word for word, omitting any names or other identifying information. All recorded interviews and typed transcripts of the interviews will be stored in a password-protected computer file on the computer in Ms. Harris' office. At the completion of the study, sociodemographic forms will be destroyed. This consent form will be filed in a locked file cabinet in Ms. Harris' office.

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Taking part in this study is your choice. There will be no penalty if you decide not to be in the study. You are free to withdraw from this research study at any time. Your choice not to participate or to withdraw from the study will not affect any services you are now receiving.

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Questions

If you have any questions, concerns, or complaints about the research, you may contact Ms. Yolanda Harris. She will be glad to answer any of your questions. Ms. Harris' number is 205-613-5872.

If you have questions about your rights as a research participant, or concerns or complaints about the research, you may contact the UAB Office of the IRB (OIRB) at (205) 934-3789 or toll free at 1-855-860-3789. Regular hours for the OIRB are 8:00 a.m. to 5:00 p.m. CT, Monday through Friday. You may also call this number in the event the research staff cannot be reached or you wish to talk to someone else.

Legal Rights

You are not waiving any of your legal rights by signing this informed consent document.

Signatures

Your signature below indicates you that you have read (or been read) the information provided above and agree to participate in this study. You will receive a copy of this signed consent form.

Signature of Participant Date

Signature of Investigator or Person Obtaining Consent Date

University of Alabama at Birmingham
AUTHORIZATION FOR USE/DISCLOSURE OF
PROTECTED HEALTH INFORMATION (PHI) FOR RESEARCH

Participant Name: _____
Research Protocol: A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers
UAB IRB Protocol Number: X161103004
Principal Investigator: Yolanda Harris
Sponsor: UAB School of Nursing

What is the purpose of this form? You are being asked to sign this form so that UAB may use and release your protected health information for research. Participation in research is voluntary. If you choose to participate in the research, you must sign this form so that your protected health information may be used for the research.

Why do the researchers want my protected health information? The researchers want to use your protected health information as part of the research protocol listed above and as described to you in the informed consent.

What protected health information do the researchers want to use? All medical information, including but not limited to information and/or records of any diagnosis or treatment of disease or condition, which may include sexually transmitted diseases (e.g., HIV, etc.) or communicable diseases, drug/alcohol dependency, etc.; all personal identifiers, including but not limited to your name, social security number, medical record number, date of birth, dates of service, etc.; any past, present, and future history, examinations, laboratory results, imaging studies and reports and treatments of whatever kind, including but not limited to drug/alcohol treatment, psychiatric/psychological treatment; financial/billing information, including but not limited to copies of your medical bills, and any other information related to or collected for use in the research protocol, regardless of whether the information was collected for research or non-research (e.g., treatment) purposes.

Who will disclose, use and/or receive my protected health information? All Individuals/entities listed in the informed consent documents, including but not limited to, the physicians, nurses and staff and others performing services related to the research (whether at UAB or elsewhere); other operating units of UAB, HSF, UAB Highlands, Children's of Alabama, Eye Foundation Hospital, and the Jefferson County Department of Health, as necessary for their operations; the IRB and its staff; the sponsor of the research and its employees and agents, including any CRO; and any outside regulatory agencies, such as the Food and Drug Administration, providing oversight or performing other legal and/or regulatory functions for which access to participant information is required.

How will my protected health information be protected once it is given to others? Your protected health information that is given to the study sponsor will remain private to the extent possible, even though the study sponsor is not required to follow the federal privacy laws. However, once your information is given to other organizations that are not required to follow federal privacy laws, we cannot assure that the information will remain protected.

How long will this Authorization last? Your authorization for the uses and disclosures described in this Authorization does not have an expiration date.

Can I cancel this Authorization? You may cancel this Authorization at any time by notifying the Principal Investigator, in writing, referencing the research protocol and IRB Protocol Number. If you cancel this Authorization, the study doctor and staff will not use any new health information for research. However, researchers may continue to use the protected health information that was provided before you cancelled your authorization.

Can I see my protected health information? You have a right to request to see your protected health information. However, to ensure the scientific integrity of the research, you will not be able to review the research information until after the research protocol has been completed.

Signature of participant: _____ Date: _____

or participant's legally authorized representative: _____ Date: _____

Printed Name of participant's representative: _____

Relationship to the participant: _____

APPENDIX S

FOLLOW-UP PHONE SCRIPT

Phone Script for Follow-Up Call

**This is the phone script for follow-up for participants that experience emotional distress

PI: *Hello. This is Yolanda Harris from CPODD. I am following up with you because you seemed emotionally upset by some of the things we discussed during your interview. How are you feeling?*

Allow the participant to answer the question.

PI: *Do you feel like you need to talk to someone else about how you are feeling*?

Allow the participant to answer the question.

If the participant says no,

PI: *If you change your mind about needing to talk with someone, contact the CPODD and we will be happy to refer you to one of our counselors at Children's Harbor at Children's of Alabama. Thank you for your time.*

If the participant says yes

PI: *We will make a referral for you to talk with a counselor in Children's Harbor at Children's of Alabama. Someone from Children's Harbor will contact you to schedule an appointment. If you have specific questions about their counseling services, you can call Counseling Service at Children's Harbor at 205-638-5229. Thank you for your time.*

After the call is ended, the PI will contact Children's Harbor counseling line to make a referral for the participant.