

USING ONLINE HEALTH COMMUNICATION TO MANAGE CHRONIC  
SORROW: MOTHERS OF CHILDREN WITH RARE DISEASE SPEAK

by

Adriana D. Glenn  
A Dissertation  
Submitted to the  
Graduate Faculty  
of  
George Mason University  
in Partial Fulfillment of  
The Requirements for the Degree  
of  
Doctor of Philosophy  
Nursing

Committee:

\_\_\_\_\_ Dr. Charlene Y. Douglas, Chair

\_\_\_\_\_ Dr. Carol Q. Urban, 1<sup>st</sup> Reader

\_\_\_\_\_ Dr. Gary Kreps, 2<sup>nd</sup> Reader

\_\_\_\_\_ Dr. Agnes Burkhard, 3<sup>rd</sup> Reader

\_\_\_\_\_ Dr. Kathy C. Richards, Assistant  
Dean, Doctoral Division and  
Research Development

\_\_\_\_\_ Dr. Thomas R. Prohaska, Dean,  
College of Health and Human  
Services

Date: \_\_\_\_\_ Spring Semester 2014  
George Mason University  
Fairfax, VA

Using Online Health Communication to Manage Chronic Sorrow: Mothers of Children  
with Rare Disease Speak

A Dissertation submitted in partial fulfillment of the requirements for the degree of  
Doctor of Philosophy at George Mason University

By

Adriana D. Glenn  
Master of Arts  
California State University, Los Angeles, 2002  
Master of Nursing  
University of California, Los Angeles, 1993  
Bachelor of Science  
Boston University, 1988

Director: Charlene Y. Douglas, Associate Professor  
School of Nursing

Spring Semester 2014  
George Mason University  
Fairfax, VA

Copyright: 2014 Adriana D. Glenn  
All Rights Reserved

## **DEDICATION**

To my late beloved daughter Avery, you were and always will be my inspiration for all that I do in this life and the next.

To my loving husband Bill, my biggest fan and supporter, I cannot imagine this life journey without you by my side, I love you.

To my wonderful sons Liam and Aidan, you both kept me going and helped me to see it is important to live life to the fullest.

My parents, the late Dr. Daniel D. Drake and Adrienne C. M. Drake without the sacrifices you made and the nurturing of my love of learning, none of this would have been possible.

## ACKNOWLEDGEMENTS

I would like to thank my family, many friends, and colleagues who have been there for me in the very worst of times and darkest days during Avery's illness and after her passing. My loving husband, Bill helped me by providing steadfast support and faith in my ability to persevere through this journey as well as a listening ear and very sage advice.

I am incredibly thankful for the support, wisdom and guidance of my committee chair, Dr. Charlene Douglas whose "reality checks" kept me focused and determined during even my most dark days/moments. I am very thankful for committee member Dr. Agnes Burkhard for her unwavering support, methods expertise and encouragement in my journey with phenomenology. I am also very appreciative of my committee members Drs., Gary Kreps and Carol Urban who were of invaluable help in providing their expertise, insights, feedback and support. Lastly, the mothers of children with Alagille syndrome whose voice was heard by me and will be heard throughout the rare disease community as we move forward to care and support families affected by this horrible disease.

## TABLE OF CONTENTS

	Page
List of Tables .....	vii
Abstract .....	viii
Chapter 1 .....	1
Background and Significance .....	1
Purpose of the Study .....	9
Research Questions .....	9
Philosophical Framework .....	10
Conceptual Framework .....	12
Conceptual and Operational Definitions of Terms .....	15
Summary .....	17
Chapter 2: Review of Literature .....	19
Chronic Sorrow .....	20
Parental Coping and Chronic Illness .....	23
Social Supports Used by Parents of Children with Chronic Illness .....	27
Online Health Communication in Management of Use by Parents of Children with Chronic Illnesses. ....	29
Chapter 3: Methodology .....	37
Hermenutic Phenomenology .....	37
Van Manen’s Human Science Approach to Phenomenology .....	38
Research Design .....	42
Population and Sample .....	43
Instruments .....	46
Data Collection Procedures .....	48
Data Analysis .....	51
Ethical Considerations .....	54
Limitations of the Study .....	55
Summary .....	56

Chapter 4: Findings .....	57
Description of Participants .....	58
Chronic Sorrow and Unpleasant Feelings .....	59
Thematic Analysis .....	64
Online Communication is Essential to a Rare Disease Community .....	65
Theme One : Connectedness .....	72
Theme Two : Online Triggers.....	81
Theme Three : Empowerment .....	87
Theme Four : Seasons of Online Communication .....	95
Integration of van Manen’s Life Worlds .....	98
Lived Space .....	99
Lived Body .....	100
Lived Time .....	101
Lived Other .....	102
Summary .....	103
 Chapter 5 : Themes, Implications and Conclusion .....	 105
Return to Research Questions and Assumptions .....	105
Themes .....	106
Connectedness .....	108
Online Triggers .....	110
Empowerment .....	112
Seasons of Online Communication .....	114
Implications .....	116
Implications for Practice .....	116
Implications for Education .....	118
Implications for Research .....	121
Implications for Policy .....	122
Conclusion .....	123
 Appendix A: Invitation and information Flyer .....	 125
Appendix B: Invitational Letter .....	126
Appendix C: Consent Form .....	127
Appendix D: Interview Guide.....	129
 References .....	 132

## LIST OF TABLES

Table	Page
Table 1: Theoretical and Operational Definitions of Study Variables .....	15
Table 2: Demographic Data of Participants.....	59
Table 3: Themes and Sub-themes.....	64



## **ABSTRACT**

### **USING ONLINE HEALTH COMMUNICATION TO MANAGE CHRONIC SORROW: MOTHERS OF CHILDREN WITH RARE DISEASE SPEAK**

Adriana D. Glenn, PhD

George Mason University, 2014

Dissertation Director: Dr. Charlene Y. Douglas

People with rare diseases share certain common psychosocial characteristics that provide opportunities for research studying these characteristics as they relate to impacting large groups of people. Similar to families coping with more prevalent chronic diseases, families dealing with rare diseases may experience common reactions upon receiving a diagnosis. The ability of those affected by rare disease to respond and manage the condition depends upon psychosocial factors such as sorrow and coping skills. Online communication has profoundly impacted the rare disease community. Using the theory of chronic sorrow as a framework, the purpose of this qualitative study was to explore the lived experience of mothers of children with Alagille syndrome in using online health communications to manage their chronic sorrow. Analysis was conducted by utilization of van Manen's (1990) interpretive hermeneutic approach to further elucidate the meaning of the mothers' experience.

Sixteen mothers of children with the rare disease Alagille syndrome were interviewed using a semi-structured interview format. The overarching theme that emerged was *Online Communication is Essential to a Rare Disease Community*. In addition, four interrelated themes were identified which provided a more complete description of the lived experiences of these mothers. Theme one, *Connectedness* captures the mothers' express need to participate in an online community. Theme two, *Online Triggers*, describes the online experiences that initiated unpleasant feelings. Theme three, *Empowerment*, reveals the value and importance in obtaining knowledge and information from online communication. Theme four, *Seasons of Online Use* emerged to describe the notable variations in online communication behaviors with regard to information seeking according to the disease trajectory.

Mothers of children with Alagille syndrome actively engage in online communication. Using online communication helped these mothers manage unpleasant feelings and chronic sorrow. However, seeking online communication also had the ability to trigger the aforementioned emotions. Implications for practice, education, research and policy were discussed in an attempt to create an awareness of the need to address and further explore psychosocial needs of the mothers within the context of online communication.

## **CHAPTER 1**

### **Background and Significance**

Almost 7,000 rare diseases collectively affect approximately 25-30 million Americans and their families. Rare (orphan) diseases are defined by the National Institutes of Health as diseases that affect fewer than 200,000 people in the United States (National Institutes of Health [NIH], 2011). It has been estimated that children comprise approximately half of those individuals affected with rare diseases (NIH, 2012). Rare diseases pose a significant problem for the health care system in terms of providing sufficient support for those people afflicted with such diseases.

Understanding the psychosocial needs of people suffering from rare diseases has not been a priority among researchers due in part to the challenges in funding research that investigates treatments and medications for such diseases. Pharmaceutical companies do not prioritize research on rare diseases since the cost to develop treatments exceeds the monetary benefit for the company (NIH, n.d.). Rare disease research is not profitable because research advances tend to impact relatively few people due to the specific issues related to each disease (Ayme, Kole, & Groft, 2008). The Rare Disease Act of 2002 led the NIH to create the Rare Diseases Clinical Research Network (RDCRN) to address the challenges of rare disease research. The RDCRN established research consortia, which allow researchers from many disciplines to share access and

resources in order to find the causes of these diseases and develop treatment therapies and medications (NIH, 2011). The RDCRN's aim to fill the void in studying causes and treatments, however, precludes extensive research on psychosocial issues that may impact individuals with rare diseases and their families.

People with rare diseases share certain common psychosocial characteristics that provide opportunities for research studying these characteristics to impact large groups of people. Similar to those coping with more prevalent chronic diseases, families dealing with rare diseases experience common reactions upon receiving a diagnosis, including shock, confusion, relief and grief/sadness (Hatton, Canam, Thorne, & Hughes, 1995). The ability of those affected by rare disease to respond and manage the condition depends upon psychosocial factors such as sorrow and coping skills.

The parental struggle with various aspects of their child's chronic illness may cause the parent to experience grief and/or sorrow (Eakes, 1995). The feelings of uncertainty surrounding the diagnosis of the chronic illness of their child coupled with their inability to achieve closure or resolution related to the grief can create chronic sorrow in parents (Eakes, 1995; Lindgren, Burke, Hainsworth, & Eakes, 1992; Vickers, 2005). Olshansky (1962) first described the term chronic sorrow to explain a normal psychological grief response that occurred in parents of children with severe mental retardation. He recognized the lifelong episodic sadness these parents experienced differed from grief models in the fact the parents' loss was cyclical and varied in intensity over the lifetime of the child with the chronic condition; these parents never achieved closure, in contrast to parents grieving the death of their child. Olshansky (1962)

essentially developed a concept that addressed grieving as never-ending loss. The chronic sorrow concept suggested that what these parents experienced was natural and that the parents learned to adapt and not accept the circumstances.

Parents caring for a child with any chronic illness generally seek and use social support. Social support encompasses supportive resources that can be emotional, tangible, and either informational or companionship (Scharer, 2005). The impact of social support on both physical and mental health has been studied extensively (Croezen, Picavet, Haveman-Nies, Verschuren, de Grot, & van'tVeer, 2012; Iyers, Chernoff, DeVet, & Young, 2001; Wang, Wu, & Liu, 2003). In general, the research supports the idea that social support contributes to improved physical and mental health of parents, making social support an important factor to consider in the management of health.

Parents of children with chronic illnesses generally seek social support from health professionals, family, friends and faith (Coffey, 2006). However, unlike more common chronic conditions, the isolating nature of rare diseases makes it more difficult for the parents to find social support to advocate for their health and welfare (Dellve, Samuelsson, Tallborn, Fasth, & Hallberg, 2006). Parents dealing with rare diseases often do not have access to health care providers who are knowledgeable about their child's condition and, thus, find it difficult to obtain health information regarding the specific condition afflicting their child. In addition, the rarity of the disease causes parents to experience geographic isolation from other parents whose children suffer from such diseases, making it difficult to form social support networks. Therefore, parents of

children affected by rare chronic disease may seek new and different means of gaining health information and support to help address psychosocial issues.

The internet has changed the way many people including people with rare diseases, obtain health care information. More adults are using the internet for health information and support (Pew Internet & American Life Project, 2012). As of May 2013, it is reported that 85% of adults in the U.S. use the internet (Pew Internet & American Life Project, 2013). When adults use the internet 72% report searching for health information with about half of the health information research being conducted on behalf of someone else (Pew Internet & American Family Life Project, 2012). According to internet survey data, women in particular are more likely to search for health related information and use online support groups (Pew Internet & American Life Project, 2007).

Adults seeking health information online have many options. Most adults begin their online search for health information with search engines like Google, Bing or Yahoo (Pew Internet & American Life Project, 2012). Search engines allow people to quickly find large amounts of information regarding their illnesses (Pew Internet & American Life Project, 2011a; Pew Internet & American Life Project, 2012). While health care providers remain the primary resource of health information particularly within the context of technical aspects of a disease, when a health situation involved more practical information or issues involved coping, then individuals (non-professionals) were the preferred source of information (Pew Internet & American Life Project, 2011b). Online support groups, chat rooms and social networking sites can all be viewed as non-traditional resources that offer social support and health information (Laskar, Sogolow, &

Sharim, 2005). Social networking sites have enjoyed substantial growth as a form of online communication particularly among older internet users. In 2005 social media use among all adults (age 18 and older) hovered in the single digits at about 8% (Pew Internet & American Life Project, 2013). Current survey data reports the use of social media adults reports a range of: 89% among ages 18-29; 78% among those ages 30-49; 60% among ages 50-64 and 43% among ages 65 and older. Overall, the data reflects an increase among all adults of 65% from 2005 (Pew Internet & American Life Project, 2013).

The use of online health communication and support by families of children with rare disease potentially helps families manage their emotions or emotional responses for several reasons. Families learn more about how diseases and their symptoms affect their children, and about the positive and negative effects of treatment and therapies aimed at curing or reducing the impact of the rare disease. In addition, families can easily find information about their child's rare condition as health care providers may not be the primary resources of information because of the rarity of the condition. Lastly, families share the daily struggles of living with a rare disease through non-traditional support groups, blogs, in-services at schools, attendance at conferences and faith communities due to the difficulty in finding the critical mass of people in one location needed for traditional types of in person support groups.

The beneficial impact of support groups on parents of children with chronic illness, regardless of the format, is well documented in the literature (Coffey, 2006; Hodges & Dibbs, 2010; Hunyard, 2009; Law, King, & Stewart, 2001). Support groups

provide parents with emotional and psychosocial support and offer them useful information about the disease and resources to help improve the quality of their lives and the lives of the children (Liu, Chao, Huang, & Chien, 2010; Solomon, Pistrang, & Barker; Trollik & Severinsson, 2005). The online support group format has allowed geographically dispersed families to share daily stresses, knowledge/experiences, and to express difficult or confusing emotions without fear of being judged (Baum, 2004). Thus, new forms of social support such as online health communication and internet support groups may be vital to assisting families of children with rare chronic disease.

Research on the disadvantages of online health communication has not been undertaken as widely as research regarding the advantages of new forms of communication. The limited analyses of the disadvantages to online health communication focus on the risk of participation in online support groups and have generated findings that are not consistent. Reported disadvantages include inappropriate/unrelated messages/postings, negative emotions, the significant volume of email and lack of physical contact (Han & Belcher, 2001). Conversely, other studies have provided evidence that lack of physical contact was not necessarily a disadvantage, but a possible advantage (Baum, 2004).

This study focuses on one rare disease, Alagille syndrome, and how new forms of communication and social support may impact the associated psychosocial consequences of the disease with regard to chronic sorrow in mothers of children with the syndrome. Alagille syndrome was first described in 1975 by the French hepatologist, Dr. Daniel Alagille (Spinner, Colliton, Crosnier, Krantz, Hadchouel, & Meunier-Rotival, 2001).



Alagille syndrome results from a rare autosomal dominant genetic disorder that impacts an important cell communication pathway. Most people with Alagille syndrome have a mutation in the Jagged1 (JAG1) gene on chromosome 20 (Spinner et al., 2001) that produces the Jagged-1 protein, which is inserted into various cell membranes. The other set of mutations that cause Alagille syndrome involves the Notch2 gene on chromosome 20, which produces the receptor for the Jagged-1 protein (Kamath, et al, 2012; McDaniell, et al, 2006). Each of these mutations impacts the ability of the Jagged-1 protein to properly transmit signals to cells regarding their development.

Alagille syndrome is a very complex disease that can affect many parts of the body. The mutations cause a reduction in the number of bile ducts in the liver, which increases accumulation of bile (ALGS National Digestive Diseases Information Clearing House [NDDIC], 2009). Greater than ninety percent of individuals who have Alagille syndrome also have congenital heart defects (McElhinney et al, 2002). Additional body systems that can be affected as a result of this rare disease include kidneys, blood vessels, eyes, face and the skeleton. Alagille syndrome symptoms generally become evident in infancy or early childhood. The estimated prevalence of Alagille syndrome is 1 in every 70,000 live births. The severity of this disease can vary widely with symptoms ranging from so mild as to go unnoticed to severe heart/liver disease requiring transplantation; sometimes the result is death. No known cure exists for Alagille syndrome. Most available treatments target improving the function of the heart and reducing the effects of impaired liver function (NDDIC, 2009).

This study of psychosocial consequences of Alagille syndrome and resources used by mothers of children with Alagille syndrome is important because these factors have the potential to affect health outcomes for the mother and the child. Alagille syndrome is a very challenging disease in terms of parental coping, but little is known with regard to how mothers of children with Alagille syndrome use online health communication to manage the sorrow associated with the diagnosis. The lack of information regarding online health communication and the experiences of sorrow has direct implications regarding the achievement of optimal health of mother and child, as well as their abilities to advocate on their child's behalf. In preparing to counsel mothers of children with this disease, nurses need to consider the very real daily experiences encountered and how mothers manage them. This study fills a gap in Alagille syndrome and rare disease research because the findings will advance our understanding of how mothers use various forms of online health communication to manage their sorrow response to a rare medical condition. In addressing this gap in the research, insights will be gained that can be used as a basis for identification and guidance of social support for mothers of children with Alagille syndrome. In addition, the findings will help in designing both interventions that address the unique needs of mothers with Alagille syndrome children and other interventions with more general applicability to those with other rare diseases (as well as perhaps those with more prevalent chronic diseases) due to the above discussed psychosocial commonalities across rare diseases.

## **Purpose of the Study**

The purpose of this study is to explore the lived experience of mothers of children with Alagille syndrome in using online health communications to manage their chronic sorrow. The isolating nature of rare disease in conjunction with health care providers' limited knowledge about specific rare diseases and lack of support to families may influence a mother's experiences with online health communication. We must develop studies describing factors having potential implications on the psychosocial health of mothers, as they often are the primary caregiver and advocate for their child. Research identifying how these mothers' experiences differ from those of mothers of children with more common chronic illnesses may lead to heightened awareness among health care providers in the subtlety of plans of care, changes in how online health communication is incorporated in to health care and further research that may contribute to efficient and effective interventions.

## **Research Questions**

Main Research Question for Consideration: What is the lived experience of mothers of children with Alagille syndrome using online health communications to manage their chronic sorrow?

Additional research questions:

- What chronic sorrow experiences are reported by mothers of children with Alagille syndrome?

- What forms of online health communication are used by mothers of children with Alagille syndrome and what are their experiences with online health communication?

### **Philosophical Framework**

Alagille syndrome is a very challenging rare disease of which little is known with regard to how mothers successfully manage the sorrow they may feel and ultimately learn to advocate for their child. The need to understand the daily experiences of mothers of children with Alagille syndrome and how they manage chronic sorrow by the use of online health communication is critical for nurses to recognize as they help and counsel mothers dealing with this disease and other rare diseases. Phenomenological inquiry was selected as the research method because it allows the researcher to obtain rich and detailed understanding of the phenomenon or problem of interest (Creswell, 2007). This is achieved by describing individual experiences and identifying phenomenon common to those individuals' reported experiences (Creswell, 2007). According to Streubert and Carpenter (2011), topics suitable for a phenomenological design include "those central to humans' life experiences (p.88)".

Phenomenology is described in the literature as both a philosophy and a method. In order to use phenomenology as a method, the researcher needs to have a grasp on the philosophic tradition he/she will use to understand the human experience. The aim of phenomenology as a method of research is to come to know the daily lived experiences of human beings by exploring and identifying the subtle, hidden or subconscious meanings within the context of those experiences; it describes the essence of a

phenomenon (Rose, Beeby, & Parker, 1995). This aim can be met by using either hermeneutic phenomenology or transcendental phenomenology as an approach (Creswell, 2007).

In general, hermeneutic phenomenology researchers describe and interpret processes as related to the nature of the lived experience, while transcendental phenomenology researchers place a greater emphasis on describing the experiences of the participants (Creswell, 2007). In addition, a significant differentiating feature between these approaches appears to be the hermeneutic tradition does not require bracketing (Lowes & Prowse, 2001). In bracketing or epoché, the researcher attempts set aside preconceptions and personal experiences. Husserl developed the epoché concept during the German phase of the phenomenological movement (Creswell, 2007). However, it is notable that processes called “decentering” advocated by Munhall (2012) are approaches specific to the hermeneutic tradition that are somewhat similar to bracketing. Decentering attempts to provide the researcher with an awareness of his/her preconceived beliefs, values and ideas (Munhall, 2012).

Heidegger, another prominent leader of the phenomenological movement during the German phase, expanded on Husserl’s work and embraced the interpretive elements of the hermeneutic view (Creswell, 2007; Streubert & Carpenter, 2011). Hermeneutic philosophy requires an interpretation of the phenomenon that supports reflecting, refocusing and further understanding; it is fluid and dynamic (Allen & Jenson, 1990). Decentering processes aid the researcher in acknowledging personal experiences that could affect his/her interpretation of a phenomenon, yet allows him/her to retain an

emic perspective. This study uses the phenomenology method of interpretation as outlined by van Manen (1990). However, practical applications of Munhall's (2012) work include decentering. The researcher used a decentering technique with van Manen's procedures to develop a more faithful or precise description of the phenomenon within the context of the hermeneutic tradition as first defined by Heidegger (Streubert & Carpenter, 2011). The researcher felt this approach was necessary to use because of her experiences as a mother of a child with Alagille syndrome.

### **Conceptual Framework**

The psychosocial impact of caring for a child with a chronic illness has been widely studied (Meleski, 2002; Stepney, Kame, & Bruzzese, 2011). Parents caring for a child with a chronic illness experience greater psychological and emotional distress than parents of healthy children (Isaacs & Sewell, 2003). The literature shows that parents experience a range of emotions, similar to grief reactions, when caring for a chronically ill child. Parents' emotional responses to their child's diagnosis include: shock, disbelief, denial, anger and feelings of guilt, decreased self-worth and decreased confidence (Eakes, 1995; Vickers, 2005). These emotions are due to the psychological stressors and unpredictability in the course of their child's illness (Dellve, Samuelsson, & Tallborn, 2006). However, unlike grief theories, which are linear, time-bound and culminate with acceptance of the loss (Scornaienchi, 2003), parents of children with chronic illness experience a recurring disparity between their expectations for their child and the reality of the constraints imposed by the disease or disability (Eakes, Burke, & Hainsworth, 1998). Moreover, they must respond to an ongoing loss with periodic

triggering events (Eakes, 1995; Northington, 2000; Vickers, 2005). This emotional response to ongoing loss is described as the concept of chronic sorrow (Eakes, Burke, & Hainsworth, 1998).

Concept analyses of chronic sorrow have been conducted by Teel (1991) and Lindgren et al. (1992) to differentiate its features and characteristics from other emotional and psychological responses like depression. Teel (1991) found that people dealing with recurring issues involving loss adapt differently than people who deal with a singular catastrophic event involving loss, such as the death of a loved one. Grief theories describe stages individuals progress through when dealing with a loss such as death. The key point of grief theories is eventual acceptance of the situation in addition to adaptation of the circumstances. When an individual is not able to accept the loss, prolonged grief is a pathological result. In contrast to grief models, chronic sorrow is a way to understand situations where an ongoing living loss is experienced and feelings of grief or sadness are triggered periodically. Similar to grief theories, the individual has to learn to adapt to the situation; however, the individual does not accept the situation because there is no finality. Teel (1991) found chronic sorrow to be a normal response to an ongoing loss. The onset of chronic sorrow was “precipitated by the permanent loss (exclusive of death) of a significant relationship” (Wikler 1981 as cited by Teel, p. 1316). Teel (1991) ultimately expanded upon Olshansky’s term and defined chronic sorrow as “a pervasive psychic pain and sadness, stimulated by certain trigger events, which follows loss of a relationship of attachment” (p. 1316).

Lindgren et al. (1992) described the differences between chronic sorrow and depression offering further clarification of and insight into the concept. Unlike chronic sorrow, depression may not have a precipitating loss that accompanies the symptoms. Furthermore, chronic sorrow is a reaction to multiple losses occurring over time, is periodic, permanent and does not interfere with daily functioning.

The middle range nursing theory of chronic sorrow helps researchers understand and frame their work in studying a natural response to loss. Eakes, Burke and Hainsworth's (1998) foundation of the theory of chronic sorrow is based on the premise that an ongoing disparity results from the loss experience, which creates chronic sorrow. Chronic sorrow is cyclical and characterized as "pervasive, permanent, periodic and potentially progressive in nature" (Eakes, Burke, & Hainsworth, 1998, p. 180). Chronic sorrow can occur in an individual affected with a chronic illness and/or the caregiver of such an individual. The model includes: antecedents, trigger events and internal/external management methods (Eakes, Burke, & Hainsworth, 1998).

In the case of parents of a chronically ill child, the antecedent to the experience of chronic sorrow most often occurs at the diagnosis of the child's illness. Trigger events may include developmental milestones, special events, and certain activities or situations that reflect the disparity between the "ideal" and the reality with their chronically ill child (Eakes, Burke, & Hainsworth, 1998). The theory of chronic sorrow, though not widely used in the literature, has been applied to a variety of populations, including children and adults with cognitive and physical impairments as well as chronic illnesses (Bowes, Lowes, Warner, & Gregory, 2009; Eakes, 1995; Mallow & Bechtal, 1999; Northington,



2000; Scornaienchi, 2003). The theory may help health care professionals recognize chronic sorrow as a normal reaction mothers' experience as they adjust to life with a child who has a rare chronic disease. The metaphor of the spotlight describes the use of theory in this study: "*Theory is a spotlight. It illuminates what you see*" (Maxwell, 2013, p. 49). Thus, the theoretical lens derives from prior research in chronic sorrow and is helpful for the purpose of forming the research questions, defining terms, and identifying issues/questions and relationships that are important to examine with regard to chronic sorrow.

### **Conceptual and Operational Definitions of Terms**

The variables considered in this study are defined in Table 1. The terms are defined in the table below.

Table 1. Theoretical and Operational Definitions of Study Variables

<b>Variable</b>	<b>Theoretical Definition</b>	<b>Operational Definition</b>
Chronic Sorrow	Cyclical sorrow/sadness experienced by individuals/caregivers that is "pervasive, permanent, periodic and potentially progressive in nature" (Eakes, Burke, & Hainsworth, 1998, p. 180.	Self-reported feelings of sadness by the mother related to her child's diagnosis of Alagille syndrome as assessed by the interview guide based upon Burke/Nursing Consortium Research on Chronic Sorrow (NCRCS) Questionnaire-Caregiver Version (Burke, 1989; Eakes, 1995)
Delayed/Atypical Developmental Milestones (child's)	Skills acquired "by a developing child, which should be achieved by a given age; failure to	Child with delayed or atypical development in at least one of the following areas: physical, cognitive, communication, social/emotional or adaptive behaviors as

	achieve a particular milestone by a given age is indicative of developmental delay” (A Dictionary of Nursing, 2008)	reported by the mother during the interview.
Illness Trajectory	The extent/degree of uncertainty with which the illness manifests; it includes both immediate and long term prognoses (Ogden, 1999).	Child’s course of illness related to Alagille syndrome that includes: initial and additional diagnoses, procedures, hospitalizations, medications etc. behaviors as reported by the mother on the demographic survey. Examples of questions: What diagnoses does your child have and what was the child’s age at diagnosis?
Online Health Communication (e-Health)	e-Health (online health communication) is “the use of digital information and communication technologies to improve people’s health and health care” (U.S. Department of Health and Human Services, 2012).	Self-reported use of internet by mother for health related information on her child’s condition. Online health communication include: list-serves, online bulletin boards, chat rooms, support group websites (sponsored by health care organizations, individuals or advocacy groups), Facebook, Twitter, blogs and other online communication/forms of technology involving the use of the internet as reported by the mother on the demographic survey. Examples of questions: Do you use the internet for health information? How often? Do you have accounts for Facebook? Twitter? How often do you use social media for health related information about your child? Do you maintain a blog related to your experiences caring for your child with Alagille syndrome?
Psychosocial Stress	Psychosocial is defined as “pertaining to a combination of psychologic and social factors” (Mosby’s Medical Dictionary,	Self-report of psychological/emotional reactions experienced when the mother perceives a situation(s) requiring resources beyond her immediate coping abilities as assessed by the interview guide based upon Burke/NCRCS Questionnaire-Caregiver

	2009). Stress is defined as an emotional experience associated with “predictable biochemical, physiological and behavioral changes” (American Psychological Association, 2012)	Version (Burke, 1989; Eakes, 1995). Any event reported by the mother that results in the mother’s self-reported feelings of heightened sadness related to her child’s diagnosis with Alagille syndrome which includes delays in developmental milestones and/or illness trajectory as assessed by the interview guide based upon Burke/NCRC Questionnaire-Caregiver Version (Burke, 1989; Eakes, 1995).
Trigger Events	Trigger events are those occurrences that reflect the disparity between the “ideal” and the reality with their chronically ill child and may include developmental milestones, special events, and certain activities or situations (Eakes, Burke, & Hainsworth, 1998)	Any event reported by the mother during the interview that results in the mother’s self-reported feelings of heightened sadness related to her child’s diagnosis with Alagille syndrome which includes delays in developmental milestones, circumstances, and/or illness trajectory.

---

### **Summary**

Collectively, rare diseases affect many Americans, with approximately half of the affected individuals being children. The ability of mothers who care for children afflicted by rare diseases to adequately respond and manage the condition is impacted by psychosocial variables. These mothers may experience chronic grief and/or sorrow, as is often noted in parents of children with chronic illness.

Parents usually seek and use social support when caring for their child with a chronic illness. However, the isolating nature of rare diseases makes it difficult for mothers to use social support from the traditional first line sources that are available to mothers whose children have more common chronic illnesses. The challenge of managing the rare disease coupled with the chronic sorrow experienced by the parent(s) prompts many parents to seek social support from the internet; this is known as e-Health or online health communication. Online health communication encompasses a wide variety of digital information and communication technologies. The emerging research in online communication currently supports the importance and viability of this type of social support for individuals managing many types of chronic illnesses.

This study focuses on mothers caring for a child with the rare disease called Alagille syndrome. Using the theory of chronic sorrow as a guide, this study seeks to increase the understanding of how these mothers use online health communication to manage their chronic sorrow in response to a chronic illness that is rare. This knowledge will help nurses guide mothers to online sources and enable nurses to better understand, advocate for and develop improved supports and potentially guide the development of interventions.

## CHAPTER 2

### Review of Literature

The review of literature explores factors related to parental coping, social supports and online communication in parents of children with chronic illness, with an emphasis on mothers caring for a child with a rare chronic disease. The literature reviewed reflects a variety of health disciplines. This review of the literature will proceed in four parts. The first section will address the psychological consequences of chronic sorrow in parents of children who have chronic conditions. The second section addresses parental coping with regard to caring for a child with a chronic illness. The third part will address the typical social supports used by parents of children with chronic illness. The last part discusses the literature as it relates to online health communication for parents of children with special health care needs.

The search for related literature was conducted using the CINHALL, Medline, ProQuest, Evidence Based Medicine Reviews and Psych-Info databases. The keywords used for the search include: *Alagille syndrome, chronic sorrow, chronic illness, children, coping, electronic communication, family, internet, mothers, online, orphan diseases, parents, rare diseases, self-help groups, social support, support groups.*

## **Chronic Sorrow**

The term chronic sorrow was first described by Olshansky (1962) to explain a normal psychological grief response that occurred in parents of children with severe mental retardation. Central to understanding Olshansky's (1962) concept is the parental experience is not one of accepting the circumstances involving their child, but adapting to those circumstances. In general, the body of knowledge addressing chronic sorrow as defined by Olshansky (1962) and further clarified by Teel (1991) and Lindgren (1992) is limited.

The majority of the limited research on chronic sorrow used qualitative methodologies with small numbers of participants. However, this small body of literature addresses chronic sorrow in a variety of adult and pediatric populations. In adult populations chronic sorrow has been described in individuals with cancer (Eakes, 1993; Rosenberg, 1998), multiple sclerosis (Ahlstrom, 2007; Hainsworth, 1994; Hainsworth, 1996; Isaksson & Ahlstrom, 2008; Isaksson, Gunnarsson, & Ahlstrom, 2007; Liedstrom, Isaksson, & Ahlstrom, 2008), Parkinson's disease (Lindgren, 1996), substance abusing females who were victims of child abuse (Smith, 2009), and HIV positive individuals (Lichtenstein, Laska, & Clair, 2002).

In the pediatric population, the research largely addresses parental experiences regarding chronic sorrow in caring for their child with a mental or physical illness that is chronic in nature. Chronic sorrow has been identified in parents of children with mental retardation (Damrosch & Perry, 1989), neural tube defects (Hobdell, 2004; Hobdell & Deatrick, 1996) and chronic illnesses such as Type 1 diabetes, epilepsy and sickle cell

disease (Bowes, Lowes, Warner, & Gregory, 2008; Hobdell et al., 2007; Northington, 2000). In addition, case studies of mothers of children with two rare conditions (unspecified neurological disorder and lissencephaly) where chronic sorrow was identified were also reported in the literature (Bettle & Latimer, 2009; Scornaienchi, 2003).

In general, the research on pediatric populations via the parental perspective supports the importance of coping and social support as it relates to chronic sorrow. Gender differences in chronic sorrow were noted by Damrosch and Perry (1989) who studied parents of children with Down syndrome. In comparing mothers' and fathers' patterns of adjustment and chronic sorrow frequency and coping, they found fathers reported adjustment to the diagnosis as a slow, gradual recovery. Mothers, however, described adjustment patterns of peaks and valleys with occasional crises.

Parents have also reported that chronic sorrow is not viewed as a normal response by health care providers. Large scale qualitative research findings illustrated the lack of support or empathy parents experienced from health care professionals (Bowes, Lowes, Warner, & Gregory, 2009; Eakes, 1995; Griffin & Kearney, 2001). In addition, in two smaller case studies, mothers described health care providers as viewing the parent's optimism, hope, and advocacy as denial of their reality (Bettle & Latimer, 2009; Scornaienchi, 2003).

Parents need strength and support to cope with the attitudes of many health care providers. Parents and caregivers of children reported a sense of joy and strength comes from their child that helps them deal with chronic sorrow (Northington, 2000;

Scornaienchi, 2003). Also, in dealing with the chronic sorrow, parents recognized the need for emotional support and sought various sources of social support (Bowes, Lowes, Warner, & Gregory, 2009; Northington, 2000).

Eakes' (1995) study of parents of adult children with chronic mental illness yielded results consistent with some of the findings in studies of parents of younger children. However, in her small qualitative study, she noted Black parents did not experience chronic sorrow. While results need to be interpreted with caution, this finding does raise the question as to whether culture or family structure is associated with chronic sorrow.

Quantitative research addressing chronic sorrow is rare. Two studies by Hobdell were identified that used a quantitative approach to study chronic sorrow. Hobdell (2004) studied parental chronic sorrow in 132 parents (63 mother/father pairs and 6 single parents) following the birth of a child with a neural tube defect. She identified significant differences between genders and suggested that there is an association between depression and chronic sorrow. Hobdell and colleagues (2007) later studied chronic sorrow and coping in families of children with epilepsy using the Adapted Burke Questionnaire and the Coping Health Inventory for Parents (CHIP). They found a significant relationship between the grief component of sorrow (feelings of grief) and Coping Pattern II of the CHIP, which addresses maintaining social support, self-esteem and psychological stability.

The literature on chronic sorrow continues to evolve as researchers study the concept in a wider range of populations. Studies on chronic sorrow have traditionally



used qualitative approaches involving small sample sizes. The strength of this small body of research lies in the depth of meaning and clarification of the concept of chronic sorrow. Furthermore, it provides evidence of the existence of chronic sorrow in parents of children with chronic conditions, in addition to the range of other emotions commonly experienced by these parents. However, the research base could be strengthened by further studies designed to measure the level of chronic sorrow, the examination of additional variables and their association with chronic sorrow, and the use of larger samples.

### **Parental Coping and Chronic Illness**

The term coping appears more frequently in the literature than chronic sorrow. Based upon the literature, it is apparent that individuals must incorporate coping strategies in order to deal with the situation and manage the accompanying stresses. Coping in the context of psychological perspective has been defined by Lazarus and Folkman (1984) as “constantly changing cognitive and behavioral efforts to manage specific external and internal demands that are appraised as taxing or exceeding the resources of a person” (Lazarus & Folkman, 1984, p. 141).

In general, coping refers to adaptive or constructive behaviors and can be classified into three types: 1) appraisal focused; 2) problem focused; and 3) emotion focused (Weiten, Dunn, & Hammer, 2011). Appraisal focused behaviors involve changes in the way a problem is viewed, such as changing goals or distancing oneself from the problem (Weiten, Dunn, & Hammer, 2011). Problem focused strategies address the need to deal with the actual problem, which is achieved by seeking more information

about the problem and learning new skills to manage the problem (Weiten, Dunn, & Hammer, 2011). Appraisal and problem focused coping are also described as action strategies and cognitive coping (Eakes, Burke, & Hainsworth, 1998). Emotion focused coping behaviors encompass management of emotions resulting from the problem including distraction and working through anger (Weiten, Dunn, & Hammer, 2011). Emotion focused coping can involve positive elements such a crying and embracing spirituality practices (Allen & Marshall, 2010; Eakes, Burke, & Hainsworth, 1998). However, sometimes emotion focused coping can result in maladaptive coping that could potentially result in mental illness (Weiten, Dunn, & Hammer, 2011). Most individuals use more than one type of coping method (Canam, 1993; Lazarus & Folkman, 1984).

The literature related to parental coping in the context of care of a chronically ill child is studied more extensively in mothers. However, more recent studies have focused on identifying differences in coping behaviors between mothers and fathers, and exploring coping behaviors that are associated with successful adaptation to the stressor/circumstances (Brazil & Krueger, 2002; Hodgkinson & Lester, 2002; Katz, 2002; Tucker, Butler, Loyuk, Desmond, & Surrency, 2009).

Research on mothers and coping behaviors show that mothers tend to use appraisal and problem focused strategies to successfully adapt to caring for a chronically ill child (Brazil & Krueger, 2002; Hodgkinson & Lester, 2002; Tucker, Butler, Loyuk, Desmond, & Surrency, 2009). A metasynthesis of qualitative literature on parenting a child with chronic illness yielded seven themes from 11 studies (Coffey, 2006). The majority of the participants in the 11 studies were analyzed were mothers. The theme

taking charge encompassed “tenacious information seeking” (p.58) and offers further evidence of the use of problem focused coping in adaptation of the care of their chronically ill child (Coffey, 2006). The specific problem focused coping behavior that appears most helpful is seeking support from others (Hodgkinson & Lester, 2002).

Research on coping behavior differences between mothers and fathers identified emotion focused coping and its association with social support and its being a predictor for parental stress. For example, Katz (2002) developed a causal model addressing differences between mothers and fathers in adapting to a child’s chronic illness. She studied 80 mothers and 80 fathers of children who were diagnosed with life-threatening (LT) and non-life threatening chronic illness (NLT). Katz (2002) found emotional coping behavior was used the least by both mothers and fathers of both LT and NLT groups. Additionally, the parent of a child diagnosed with LT chronic illness had a greater range of coping behaviors available to use. Katz (2002) also reported both mothers and fathers use of emotion focused coping was influenced by the perception of social support received and self-esteem. An interesting finding of this study that contradicted previous research was that Katz (2002) found the influence of social support on the use of coping behaviors in fathers played a greater role than with mothers.

Dabrowska and Pisula (2010) studied stress and coping in Polish mothers and fathers of children with autism, Down syndrome and typically developing children. They found emotion focused coping in the parents of the cognitively disabled children as predictor for parental stress. The parents of the typically developing children were found to use task oriented coping (problem focused coping) and experienced lower parental

stress. This finding is consistent with the literature that illustrates problem focused coping is associated with the parents' adaptation to the child's chronic illness (Tucker, Butler, Loyuk, Desmond, & Surrency, 2009).

Limited amounts of research focuses specifically on coping in fathers of chronically ill children. The literature base on fathers' coping strategies largely derives from studies involving both the mother and father as a couple. In recent years, there has been an increased focus on describing how fathers cope with caring for a chronically ill child. In general, a father's ability to cope is associated with his concerns, the perceived concerns of the mother (Hovey, 2005; McGrath & Chesler, 2004) and the level of family support (e.g. financial and emotional) that he experienced (Hovey, 2006; Perrin, Lewkowicz, & Young, 2000). Two studies provided evidence of specific coping techniques used by fathers. Broger and Zeni (2011) and Katz (2002) reported fathers used emotion focused coping strategies. However, Broger and Zeni's (2011) descriptive research design offered greater detail about the relationship between demographic data and the coping method used by fathers. In the convenience sample of 54 fathers, they found: 1) emotion focused coping used by fathers was associated with a religious component; 2) married and older fathers used positive coping more often than younger and unmarried fathers and; 3) lower income fathers were more likely to cope by seeking social support (problem focused coping), distancing (appraisal focused coping) as well as maladaptive coping techniques of escape and avoidance.

There are many instruments available to assess coping in parents of chronically ill children. Examples of tools used to assess parental coping of chronically children

include: Burke/NCRLS Chronic Sorrow Questionnaire (Caregiver Version) (Burke, 1989; as cited in Eakes, 1995); Chronicity Impact and Coping Instrument (CICI:PQ) (Hymovich, 1984); Coping Health Inventory for Parents (CHIP) (McCubbin, McCubbin, Nevin, & Cauble, 1981); and Ways of Coping-Revised (Folkman & Lazarus, 1985). These instruments reflect the many dimensions of coping and the circumstances that can impact parents' ability to cope. The variety of instruments available make the concept of coping an attractive measure of parental/family functioning in response to a stressor such as chronic illness.

The interrelatedness of coping and chronic sorrow is well documented in the literature. Investigations focused on coping behaviors are more prevalent in the literature, which may be due to a wide range of instruments available to researchers to measure the concept. The literature on coping behaviors shows the need for emotional support and the effectiveness of social support in the facilitation of positive coping and adaptation in parents of children who have a chronic illness. The differences between mothers and fathers in their coping styles has been described, but is inconclusive with regard to how best to address the psychosocial needs of these parents as it relates to gender differences in coping.

### **Social Supports Used by Parents of Children with Chronic Illness**

The term social support describes a range of activities that stems from an individual's social network. Social support network literature generally identifies four types of social support: 1) emotional; 2) informational; 3) appraisal; and 4) material/instrumental (Birch, 1998; Eastin & LaRose, 2004). Emotional support is best

described as interaction with others that creates an environment of caring and positive emotion (Birch, 1998; Eastin & LaRose, 2004). Informational support entails sharing knowledge or assisting the individual in obtaining the needed information (Birch, 1998). Appraisal support involves providing affirmation or validation to an individual (Birch, 1998; Eastin & LaRose, 2004). Material/instrumental support is described as physically helping an individual with tangible materials (Birch, 1998; Eastin & LaRose, 2004).

Parents of children with chronic illness have consistently identified the need for social support in the forms of emotional, informational and material/instrumental support. Many parents of children with a chronic illness experience social isolation resulting from reduced time for social activities and loss of social support networks (Patterson, 2002). In addition, the family's ability to interact successfully with the community can be impacted by societal trends such as family relocation and a decrease in family size (Hanson, Kaakinen & Gedaly-Duff, 2005). Walsh (2006) reports that healthy families recognize when they need help and are more likely to seek assistance from family, friends, neighbors or community and/or counseling services. One way parents of children with a chronic illness seek assistance is through the use of support groups.

A support group is a forum where people can share common experiences/concerns, exchange knowledge and express emotions/feelings (Graziosi, 2010). Support groups can range in structure from an informal gathering of families to groups sponsored by an organization. Support groups have been recognized as an effective intervention for individuals/families caring for a loved one with special health care needs (Chien, Chan, Morrissey, & Thompson, 2005; Liu, Chao, Huang, Wei, &

Chien, 2010; Solomon, Pistrang & Barker, 2001; Trollvik & Severinsson, 2005). Support groups provide parents with emotional and psychosocial support and useful information about disease, treatment, and resources (Liu, Chao, Huang, Wei, & Chien, 2010; Solomon, Pistrang, & Barker, 2001; Trollvik & Severinsson, 2005). Support groups have been shown to facilitate parental coping ability in those families caring for children with special health care needs (Ainbender, et al., 1998). Major barriers identified with regard to participation in face to face support groups include issues regarding child care (Bouchard, 1998), time constraints, and location (Bragadottir, 2008; Hall & Irvine, 2008).

### **Online Health Communication Used by Parents of Children with Chronic Illness**

**Online health information resource behaviors of parents.** Parents of children with chronic health conditions use various online resources for health communication. These formats include: blogs, social networking sites, discussion boards, websites and emerging technologies like Skype and Google chat. In general, people tend to use online health information to: 1) find information about a specific disease before or after the doctor visit, 2) make health care related decisions, and 3) share their online findings with their physician when discussing their health concerns (Morahan-Martin, 2004). Current research on online health communication resources specifically used by parents includes the need to: supplement information from physicians, develop knowledge in order to enhance advocacy for their child and to develop social support networks (Coulson & Greenwood, 2011; Gundersen, 2011; Leonard et al., 2004; Roche & Skinner, 2009; Semere et al., 2003; Tozzi et al., 2013).

Research, though limited, has begun to address how parents use the internet for medical information. In general, the literature shows that parents turn to the internet for additional medical information, especially during stressful events like a new diagnosis, emergence of a new symptom or for a pending procedure/surgery for their child (Roche & Skinner, 2009; Semere et al., 2003). Roche and Skinner (2009) note that parents of children with a confirmed genetic diagnosis had multiple informational needs which included anticipatory guidance with regard to clinic visits and presentation of complex genetic information and sought information to be better prepared for a discussion during the visit. Parents seeking additional health information regarding their child enjoy the convenience, amount of available information and anonymity the internet offers. Consequently, the information parents obtain often influences the medical decisions they make on their child's behalf (Semere et al., 2003). However, Semere and colleagues (2003) cautioned that while parents engage in frequent internet use as a source for health information and decision making, the researchers identified significant concerns with regard to how proficient parents have truly become at locating reliable sources of health information.

Parents of children with more prevalent chronic illnesses use the internet differently than parent of children with rare diseases. The research literature has begun to differentiate between these situations. Two studies of more common chronic and potentially life-threatening pediatric conditions highlight this emerging difference. Gage and Panagaskis (2011), and Nordfelt, Angarne-Lindberg, Norwall and Krevers (2013) found that their parent sample populations did not readily turn to the internet for medical



or health related information, preferring to receive their information from a trusted health care provider. Gage and Panagaskis' (2011) mixed method study of 41 parents of children with pediatric cancer reported not seeking health information online due to: fear, concerns about accuracy of information, worries of being overloaded by the amount of information available, and because their child's oncologist told them not to go online. The parent's perceptions of online pediatric cancer information were it was "untrustworthy and frightening" (Gage & Panagaskis, 2011, p. 455). Nordfelt and colleagues (2013) described that Swedish parents of adolescents with type 1 diabetes mellitus (T1DM) retrieved information based upon their perception of: the present situation, seriousness of the present situation, previous experiences/knowledge about sources and the level of trust in the source; in general, online information reported by these parents garnered a low level of trust. These parents also described receiving information from the health care providers as being essential due in part to "the complexity and uniqueness of situations involving T1DM" (Nordfeldt et al., 2013, p. 6).

Parents of children with rare diseases also deal with complex and unique situations involving their child's health. However, parents of children with rare diseases find it difficult to obtain adequate medical information because health care professionals often have little knowledge or information to share with the parents (Leonard et al., 2004). Consequently, parents of children with rare diseases often turn to the internet first to gain direct access to information related to genetic databases, genetic disorders, diagnostic criteria, online social networks on specific genetic conditions, and other medical information resources (Schaffer, Kuczynski, & Skinner, 2008; Skinner and

Schaffer, 2006). An internet profile that emerged from 516 Italian parents of children with rare diseases, who responded to a web based survey, found these parents to be active internet users who were strongly engaged in information seeking, with two thirds likely to discuss their findings with their child's physician (Tozzi et al., 2013). Parents of children with rare conditions can experience information overload and additional anxiety from seeking health information on the internet, similar to experiences reported in parents of chronically ill children with more common disorders. However, parents of children with rare diseases report the sense of obligation and need to find the best care for their child outweighs the aforementioned risks of internet use (Schaffer et al., 2008; Skinner and Schaffer, 2006).

In general, the changing landscape of health care has led to a decrease in patient trust of their physicians (Boyer & Lutfey, 2010; Timmermans & Oh, 2010) and an increase in patient's self advocacy during visits with their health care providers (Boyer & Lutfey, 2010). The increase in advocacy leads to patients feeling more empowered. The patients' feeling of empowerment can be linked to the increased information available because of access to the internet (Boyer & Lutfey, 2010). Parents use the internet to develop knowledge of their child's condition. Knowledge of their child's disease is an important factor in the ability for the parent to regain control and to be able to advocate on behalf of their child when dealing with medical professionals (Porter & Edirippulunge, 2007). Only recently has research emerged that expands our knowledge of how parents' acquisition of health care knowledge about their child's health condition through the

internet is critical in helping parents understand and adjust to a stressful diagnosis (Gundersen, 2011).

Parents of children with chronic health conditions use online communication to seek health related information in a variety of ways and for several purposes. The parents' need for using online communication seems to vary by their child's situation (e.g., new diagnosis, upcoming procedure/surgery, etc.), the prevalence of the condition, and need to develop knowledge/expertise in the case of a child with a rare disease, so the parent can advocate on behalf of the child for best possible outcomes. While research remains scant with regard to understanding how parents evaluate and use online health communication to manage their child's chronic condition, emerging research is beginning to differentiate features that are unique to parents of children with rare diseases.

**Online support groups.** The internet provides a medium that addresses the aforementioned barriers to traditional face-to-face support group meetings because online forums are available at all times and can be accessed from virtually any location. A large number of internet or online support groups (OLSG) are available; an internet search yielded thousands of such groups. Research conducted on the effectiveness of OLSGs is limited, while research involving how parents of children with chronic illness use OLSGs is even rarer.

In general, the available research shows that OLSGs appear to be a beneficial intervention for parents of children with chronic illness (Baum, 2004; Huws, Jones & Ingledew, 2001). OLSG research remains largely exploratory, focusing on describing and identifying factors that benefited parents who participated in this type of forum. The

results indicate that mothers participate in OLSGs more often than fathers (Baum, 2004; Bragadottir, 2008; Hall & Irvine, 2008; Han & Belcher, 2001; Sullivan, 2008; Svavarsdottir & Rayens, 2003). This finding is consistent with the internet survey data showing that women are more likely to search for health related information and use support group websites (Pew Internet & American Life Project, 2007).

Some clear advantages of OLSGs cited consistently in the literature include accessibility, selective participation, anonymity, and privacy (Baum, 2004; Han & Belcher, 2001; Sullivan, 2008). OLSGs were described as helping to decrease feelings of isolation by enabling mothers to belong to a community of parents sharing similar circumstances while accounting for the problems of time, distance, and lack of child care (Han & Belcher, 2001; Sullivan, 2008). Parents who participate in OLSGs appear to experience very high levels of satisfaction stemming from factors such as receiving useful ideas, improving the caregiving relationship, and finding trust (Baum, 2004). Three studies identified empowerment and altruism as being important qualities not previously cited in the general support group literature that emerged as a result of participation in OLSGs (Baum, 2004, Leonard et al., 2004; Sullivan, 2008). Empowerment and altruism were found to be important contributors linking social support to emotional wellbeing (Baum 2004; Leonard et al., 2004; Sullivan, 2008), while altruism may be an important coping mechanism (Baum, 2004).

Recent research developments in OLSG include the role and identification of the types of support these groups offer to parents. As many as five types of OLSG support have been identified in the literature, including emotional, informational, esteem,

network, and tangible forms of support. Two studies identified OLSGs as providing high levels of emotional support (Coulson & Greenwood, 2011; Leonard et al., 2004).

Coulson and Greenwood (2011) described types of social support based upon their content analysis of three childhood cancer OLSGs. Research on the role of OLSGs and the types of support OLSGs presents a greater context through which researchers can understand how these forms of online communication help parents manage their experiences with their child's chronic illness.

Research on the disadvantages of OLSGs is not consistent and risks associated with their use have not been extensively studied. Han and Belcher (2001) identified perceived disadvantages of OLSGs, including inappropriate/unrelated messages/postings, negative emotions, significant volume of email, and lack of physical contact and proximity. Coulson and Greenwood (2011) reported some OLSG participants were disappointed by lack of responses to their messages board posts as well as the inability to communicate with group members outside of the OLSG context. However, Baum (2004) reported lack of physical contact and proximity did not appear to be an issue because OLSGs offered a greater sense of control in the amount and type of social support needed when compared to traditional face-to-face support group meetings. Baum (2004) attributed this finding to the fact that in OLSGs the in person communication modalities are bypassed. In fact, Baum (2004) found some participants actually preferred to not have physical contact and remain anonymous. Bragadottir (2008) noted another possible disadvantage of OLSGs involves how cultural considerations might impact how an OLSG is structured and facilitated, despite the option to remain anonymous.

New forms of social support such as OLSGs may be vital to improving psychosocial consequences and increasing resources. There appears to be scant literature with regard to OLSGs and their relationship to coping behaviors. Research is beginning to emerge in this area of social support, but much more is needed to establish its effectiveness as an intervention to facilitate coping. Furthermore, areas of research are needed that focus on ways to promote and evaluate the quality of OLSGs and the potential risks of participation in OLSGs.

## **CHAPTER 3**

### **Methodology**

There has been limited research about experiences of mothers of children with rare disease using online communication to manage feelings such as chronic sorrow. The researcher employed a phenomenological approach rooted in a hermeneutic orientation and guided by techniques described by van Manen to study these experiences of online communications. This chapter addresses the following methodological issues: 1) hermeneutical phenomenology; 2) research design; 3) population and sample; 4) instruments; 5) data collection procedures; 6) data analysis; 7) ethical considerations and 8) limitations of the study.

#### **Hermeneutic Phenomenology**

Phenomenology can be considered both a philosophy and a method. It is well established with regard to historical traditions and contributions to the research in various disciplines. The specific approach of hermeneutic phenomenology involves interpreting the “texts” of life and lived experiences (van Manen, 1990). The origin of hermeneutic phenomenology is most often associated with the German philosopher Martin Heidegger (Munhall, 2012). However, other European phenomenological philosophers including Paul Ricoeur (French) and Hans-Georg Gadamer (German) are also associated with this interpretative form of phenomenology (Streubert & Carpenter, 2011).

Contemporary phenomenologist and educator, Max van Manen integrates a human science perspective with phenomenology. He views phenomenology as a “philosophy of being as well as a practice” (Munhall, 2012, p. 126) and thus, is often cited in health related research. Van Manen (1990) offers guidelines in conducting hermeneutic phenomenological research, which he describes as a “dynamic interplay among six research activities” (van Manen, 1990, p. 30). The six activities are cited verbatim below:

- turning to a phenomenon which seriously interests us and commits us to the world;
- investigating experience as we live it rather than as we conceptualize it;
- reflecting on essential themes which characterize the phenomenon;
- describing the phenomenon through the art of writing and rewriting;
- maintaining a strong oriented pedagogical relation to the phenomenon;
- balancing the research context by considering parts and whole (p. 30-31).

These activities are not performed by the researcher in a linear fashion but, rather can occur simultaneously or in intervals. The six activities as applied to this study are discussed in greater detail in succeeding sections.

### **Van Manen’s Human Science Approach to Phenomenology**

**Turning to a phenomenon.** The first step in van Manen’s approach involves focusing on the nature of the lived experience. The researcher’s experiences as a mother of a child with the rare disease of Alagille syndrome facilitated the interest and desire to explore the phenomenon of the online communication and management of chronic



sorrow. Attributing importance and significance to the lived experience of women in this type of situation became the focus for phenomenological investigation “to transform this experience into a textual expression of its essence (van Manen, 1990, p. 36).

Van Manen (1990) states, “lived experiences gather hermeneutic significance as we reflectively gather them by giving memory to them” (p.37). Interpretive acts such as conversations, mediations, daydreams and inspirations help people attribute meaning to phenomenon. Thus, it was imperative that the researcher had awareness of and identified personal beliefs, biases, motives, insights, assumptions, and presuppositions related to the phenomenon under consideration. In the hermeneutic tradition, phenomenologists like van Manen typically do not advocate for bracketing one’s beliefs. Van Manen (1990) proposes researchers, “come to terms with our assumptions, not in order to forget them again, but to hold them deliberately at bay” (p.47). However, van Manen does not provide a structured way as to “come to terms” with these assumptions. The researcher sought to address concerns of potential personal biases and beliefs by incorporation of the concept of decentering.

Decentering is an approach advocated by Munhall (2012) and specific to the hermeneutic tradition. When a researcher engages in decentering, he/she attempts to create an awareness of his/her preconceived beliefs, values and ideas and consequently constructs an “unknowing” environment in which to interpret properly the lived experience (Munhall, 2012). In this study, the researcher maintained a personal journal in which she recorded thoughts associated with the phenomenon of interest for the duration of the study. Journaling occurred before interviews with the mothers and at the

conclusion of each interview. These accounts included personal reflections as well as interview details related to the study.

**Investigating the experience.** The second step requires the researcher to investigate the experience as it is lived and experienced. The researcher searches in the lifeworld for any lived experience material that might be seen as important and contributory to interpreting the meaning of lived experience (van Manen, 1990). In essence, the lived experiences are experiential data the researcher uses to explore and interpret the phenomenon. In this step, the researcher began with her personal experiences with the phenomenon by developing a personal description during a journal entry. Van Manen (1990) suggests “in drawing up personal descriptions of the lived experiences, the phenomenologist knows that one’s own experiences are also the possible experiences of others” (p.54). The researcher then used a qualitative semi-structured interview guide (Appendix D) she developed using the Theory of Chronic Sorrow as a guide. The researcher’s questionnaire encouraged a hermeneutic interview process; the mothers’ telephone interviews were the primary form of obtaining experiential data.

In phenomenology, the researcher is both a participant and instrument. Thus, she has the potential to add to the richness of data collection and analysis (Streubert & Carpenter, 2011). In addition, the hermeneutic interview “tends to turn the interviewees into participants or collaborators of the research project” (van Manen, p.63). This perspective was consistent with the aim of the researcher in using the best way to investigate the phenomenon of interest and complimented the researcher’s attempt to gather and reflect on the meaning of the lived experiences of the mothers.

**Reflecting on essential themes.** The third step in Van Manen's (1990) approach required the researcher to reflect on the data using a phenomenological lens. Van Manen (1990) states a researcher conducting human science research is involved in the "crafting of a text" (p. 78). In this step of the process, the researcher became immersed in the data in order to make the findings discussed in greater detail in Chapter 4. The researcher used the transcribed interviews, field notes and personal journal entries to develop essential themes arising from isolation of thematic statements.

**Phenomenological writing.** The fourth step of Van Manen's (1990) process of analysis involved the phenomenological writing. This is a critical step for phenomenological researchers as writing actually blends the research activity and reflection itself (van Manen, 1990). Consequently, the researcher's emerging incidental and essential themes provided structure, were analyzed and interpreted throughout the writing process within the context of four existential lifeworlds described by van Manen (1990). The lifeworlds of spatiality (lived space), corporeality (lived body), temporality (lived time) and relationality (lived human relations) are addressed in Chapter 4.

**Maintaining strong and oriented relation to phenomenon and balancing parts and the whole within context of research.** Van Manen (1990) in steps five and six instructs the phenomenological researcher in the human sciences to maintain a strong and oriented relation to the phenomenon and to balance the parts and the whole within the confines of research. He cautions researchers that it is important to maintain perspective and pragmatic application of their research findings to the lifeworld. Van Manen states, "the tendency to abstraction is a common hazard of all academic activity"

(p.138). These last two steps seek to help the researcher understand that while a hermeneutic phenomenology is interpretive and rooted in philosophy, it is a philosophy of action. The importance of being mindful of the part-whole relation of one's research is helpful in redirecting the researcher's thinking during the process of writing and interpreting. Researchers that overlook steps five and six are likely to provide inadequate interpretation of the lived experiences of participants and, thus, produce research with little practical significance.

### **Research Design**

The researcher selected a phenomenological approach for this qualitative study for several reasons. First, the main research question was best addressed by an exploration of "lived experiences" among mothers who used online health communication to manage the chronic sorrow they experienced related to their child's diagnosis of Alagille syndrome. Very limited research exists regarding the use of online communication in managing chronic sorrow. Second, very little research has been conducted regarding the psychosocial impact of rare disease; even less research has investigated the use of online health care communication by mothers of children with rare diseases. Thus, this research, which provided an in-depth description of observed phenomena, fills a void in the field. Third, research conducted in the area of online health communication has not yielded consistent findings on the potential disadvantages of online health communication in the management of illness. This study contributes to a better understanding of the disadvantages. Lastly, this design offered an optimal

approach to studying the issue because one can conduct multiple interviews to gather the rich data required to describe the phenomenon of interest.

### **Population and Sample**

The target population consisted of mothers of children diagnosed with Alagille syndrome who use online health care communication. Mothers were targeted in this study because most often the mother assumes the responsibility for managing the child's health conditions (Smith & Schaefer, 2012). Additionally, researchers indicate that mothers participate in online support groups more often than fathers (Han & Belcher, 2001; Svavarsdottir & Rayens 2003; Baum, 2004; Bragadottir, 2008; Hall & Irvine, 2008; Sullivan, 2008). Internet survey data reveal that women are more likely to search for health related information and use support group websites (Pew Internet & American Life Project, 2007). The literature on mothers' use of coping techniques shows that mothers tend to use problem focused coping, which is inclusive of information seeking behaviors that will assist the mother in learning new skills to manage her child's health issues.

A purposive sample of 16 mothers of children diagnosed with Alagille syndrome was recruited. Purposive sampling is often used in qualitative research because the researcher selects participants because they can "purposefully inform an understanding of the research problem and central phenomenon" (Creswell, p. 125).

The researcher posted an informational flyer (Appendix A) approved by the George Mason University's (GMU) Office of Research Integrity and Assurance (ORIA) on internet sites (Facebook sites dedicated to individuals with Alagille syndrome and the

Alagille Alliance website). In addition, the Alagille Alliance agreed to insert the informational flyer in the second quarter Alagille Alliance quarterly newsletter, *Links for Life*.

In addition, the researcher used snowball sampling. Snowballing, a type of purposive sampling uses one informant to find another (Streubert & Carpenter, 2011). Three of the 16 mothers were recruited using the snowballing approach. When the snowballing technique was used, the researcher asked participant mothers as well as an Alagille Alliance board member if they knew mothers who might have an interest in participating in the study. If they knew of other potential participants, the researcher requested the individual share the researcher's email contact information as well as the informational flyer (Appendix A).

All interested participants were asked to contact the researcher via telephone or internet. All participants chose to contact the researcher through the internet in email format or private message on Facebook. The researcher then emailed or sent a private message to qualified participants with a written explanation of the study that included the purpose of the study (Appendix B), as well as a review of the inclusion requirements and approximate time involved in the interview process. In addition, the researcher's reply email/message included the consent form (Appendix C) approved by the GMU ORIA.

Participants were asked to read, sign, date and return the consent to the researcher. Also, the mothers were encouraged to contact the researcher via email or phone if questions remained after reviewing the information they received. Though the consent form was sent electronically, mothers had the option of returning the consent through the

U.S. mail service; several mothers chose the latter option. After the researcher received the signed consent form, the researcher contacted the mother via email or telephone to arrange a telephone interview date and time mutually agreeable to the mother and the researcher and answered any preliminary questions.

The purposive and snowball sampling was conducted until data saturation was reached and a sample size of 16 mothers was obtained. In collecting the optimum data for study, the researcher elected to gauge an approximate sample size by using an approach integrating data saturation with sample size recommendations. Data saturation was defined by the researcher as new data and sorting confirmed categories and themes identified by the researcher. The consideration of a pre-determined sample size was specific to phenomenology and was derived from the loose guidelines offered by established qualitative researchers and qualitative studies precedents (Creswell, 2007; Mason, 2010).

The participants were required to meet the following inclusion criteria: mothers had to be English speaking, at least 18 years of age or older, and a self-identified caregiver for child/children with Alagille syndrome. A child with Alagille syndrome was defined as a child with confirmation of the disease diagnosis by a medical doctor confirmed via genetic testing and/or clinical indicators of Alagille syndrome (e.g. liver biopsy, butterfly vertebrae, posterior embryotoxin, etc.). In this study, all children were under 18 years of age.

The feasibility of the study was a bit challenging with regard to adequate recruitment, thus the need to use snowball sampling toward the end of data collection.

The Alagille community is small and geographically diverse; the combination of the internet and newsletter in recruitment proved very helpful, as did snowballing. The researcher found mothers of children with Alagille were eager to participate, but many mothers could not participate due to commitments to other children, numerous appointments with specialists for their child with Alagille syndrome, involvement in other research studies and some unfortunate/unanticipated health problems which required extended hospitalizations of their children. Two mothers who consented and were looking forward to being interviewed were unable to participate in the study due to significant/life-threatening adverse health events that occurred in their child after they agreed to participate.

### **Instruments**

The researcher developed a two part qualitative interview guide to collect data for this study (Appendix D). The first section was designed to gather basic background information on the mother and her child for purposes of describing the sample demographics. Demographic information on the mother included age, race/ethnicity, marital status, highest level of education employment and country of residence/state. Information related to the child included the age of the child with Alagille syndrome, the age when the child was diagnosed with Alagille syndrome, and other diagnoses.

The second part of the interview guide consisted of 10 open ended questions related to the mother's experiences with online communication, information seeking behaviors, and thoughts/feelings since the child was diagnosed with Alagille syndrome. The researcher drafted the questions related to the topics of online communication and



information seeking behavior. The researcher used some of the questions contained in the Burke/Nursing Consortium Research on Chronic Sorrow (NCRCS) Questionnaire-Caregiver Version to elicit the thoughts/feelings of the mothers related to the presence of chronic sorrow.

The Burke/NCRCS Questionnaire-Caregiver Version contains 16 open-ended questions that explore feelings of chronic sorrow in caregivers (Burke, 1989; Eakes, 1995). Burke developed the initial tool in 1989 to assess chronic sorrow in mothers of children with myelomeningocele disability. The Nursing Consortium Research on Chronic Sorrow (Burke, Hainsworth, Eakes, & Lingren, 1992) provided further refinements that led to the Burke/NCRCS Questionnaire-Caregiver Version becoming a tool with applicability in varied populations across the lifespan.

The Burke/NCRCS Questionnaire-Caregiver Version probes several aspects of chronic sorrow. Questions #2, #5, #8 and #16 address the occurrence of chronic sorrow, with question #9 probing for intensity of the chronic sorrow. Questions #7 and #8 are designed to elicit responses related to developmental milestones or triggers at which feelings of chronic sorrow are likely to surface. Questions #3, #4, #11 and #12 seek answers to what the caregiver found helpful or not helpful in coping with the chronic sorrow. Practical advice to other caregivers is addressed in questions #13 and #14. Lastly, the caregivers' suggestions to nurses and other caregivers are captured with participants' responses to item #15.

The researcher considered using the complete Burke/NCRCS Questionnaire-Caregiver Version. However, using the questionnaire in its entirety might have precluded

participants from sharing their lived experiences of the phenomena completely and within the context of online health communication. Thus, the researcher decided the best approach to obtain rich narrative data included integrating questions from the Burke/NCRCS Questionnaire-Caregiver Version into the researcher's qualitative interview guide that encompasses questions regarding experiences with online health communication. The interview guide was reviewed by two content experts to address possible researcher bias. Researcher bias is one of two broad validity threats often identified in relation to qualitative research (Maxwell, 2013).

### **Data Collection Procedures**

Data were collected over a 3 month period from June 2013 to August 2013. The researcher conducted individual interviews of all 16 mothers. Fifteen mothers were interviewed by telephone, while one mother elected to be interviewed using Skype. The researcher initiated the call from a private location to each mother at the mutually agreed upon date and time. The participants were reminded that their participation in the study was voluntary; they could withdraw from the study at any time and for any reason. In addition, the mothers were reminded that during the interview, they did not have to answer any interview question they did not want to answer. The researcher also asked if the participant had any questions or concerns prior to the start of the interview.

The researcher used the following interview procedure with each mother in the study:

1. The researcher assigned a pseudonym to the mother and recorded it on the interview guide. The researcher assured participants of confidentiality and reminded them of

their assigned pseudonym. The researcher asked the mother to pick a name other than the child's birth/real name that she would use to refer to her child. The researcher recorded the child's name for the study on the interview guide, along with the interview date, mode of interview (phone or Skype), and the start time of the interview.

2. The researcher introduced the semi-structured qualitative interview guide and explained that there were two parts to the interview process. The researcher told the mother that the first part was designed to gather basic background information on the mother and her child and that this part would not be audio-recorded; instead, the researcher would document the mother's responses on the form. The mother was informed the second part involved questions related to experiences with health communication, seeking information, and thoughts/feelings since the child was diagnosed with Alagille syndrome. The researcher informed the mother that the second part would be audio-recorded and that she would be put onto speaker phone so the interview could be adequately recorded.
3. The researcher completed part one of the interview guide.
4. The researcher informed the mother that the audio-recorder was going to be turned on and verified the mother's agreement and understanding that the interview now would be audio-taped.
5. The researcher placed the mother on speaker phone and verified that the sound worked.

6. The researcher turned on two digital audio-recorders located on the desk top to capture the interview. The researcher used the second audio-recorder to decrease the chances of lost data in the event of equipment malfunction or mechanical failure.
7. After a brief reminder of the type of information part two of the interview was designed to obtain, the researcher began the second part of the interview, which included the 10 questions plus probes to follow up on some of the responses. Each interview lasted from 20-75 minutes. When the interview was complete, the researcher promptly turned off both audio-recorders and noted the ending time on the interview guide. The researcher thanked the mother for her time and support.
8. The researcher reflected upon her personal beliefs, preconceptions, theories, motives, interview experiences, and personal biases in an attempt to decenter and to facilitate openness and transparency. The researcher documented these reflections in a personal journal after each interview and, on several occasions, a few days before a scheduled interview.
9. The researcher stored the informed consents, part one of the interview guide, and both digital audio-recorders in a locked cabinet. The code key linking the mothers' actual names to the pseudonym was stored in a locked file cabinet separate from the rest of the data in the study. The researcher was the only one who had access to both secured data areas. The researcher plans to destroy the transcribed data/notes/audio-recordings and audio files three years after completion of the dissertation defense.
10. The researcher personally transcribed each interviews within 72 hours of its completion.

## **Data Analysis**

The researcher analyzed data gathered from the first part of the interview to provide descriptive information about the sample of mothers. The researcher uploaded the data collected from the second part of the interview into the web application Dedoose Version 4.5 to assist the researcher in organizing, coding, and recording the themes yielded from the textual data contained in the transcripts.

The phenomenological method proposed by van Manen (1990) was used to guide the researcher in analyzing the participants' transcripts. Phenomenological themes are "the experiential structures that make up the experience" (Van Manen, 1990, p. 79). Furthermore, Van Manen described thematic analysis as the "process of recovering the theme or themes that are embodied or dramatized in the evolving meanings and imagery of the work" (p. 78). The researcher began the process of analysis by engaging in "hermeneutic phenomenological reflection" (p. 77). The researcher accomplished this by reflection and analysis of the lived experience described by the mothers, and then transitioning into conducting thematic analysis via reflection on essential themes and their meaning and significance in relation to the lived experience described by the participants.

The researcher then used van Manen's approach for isolating thematic statements. This procedure entailed the researcher first conducting a "wholistic reading" (p.93) of each transcript. The researcher engaged in this activity several times for each narrative. The wholistic reading was followed by several readings using a "selective reading approach" (p. 93) and ended with "detailed approach" (p.93) where each sentence and

sentence cluster are closely examined. This structured approach to analyzing and isolating thematic statements helped the researcher to differentiate among incidental and essential themes. After themes were identified, van Manen (1990) suggests researchers consider the themes as objects of reflection to be considered within the context of four life world existentials: lived space, lived body, lived time and lived human relation (p. 101) to guiding the researcher in interpretation.

As discussed earlier, the researcher supplemented van Manen's (1990) method of interpretation with Munhall's (2012) structured stepwise approach to qualitative research. Munhall's steps are very closely aligned with those of van Manen, but offer more detail in how best to develop phenomenological thinking. Munhall advocates for the use of decentering to aid in the task of becoming more phenomenological in one's orientation. In addition, Munhall argues "relevance must extend beyond listing essences and themes" (p. 167), so she advocates that the researcher return to the participants for validation of the descriptions of the phenomenon of interest. These two practical techniques are not identified in van Manen's work, but are very useful and complement his approach. The use of these two techniques proved critical for the researcher because of her personal experiences with the phenomenon of interest.

The researcher completed the last step of van Manen's approach to interpretation of the data, "hermeneutic phenomenological writing" (p.111). This step, at its core, requires an awareness of the subtleties of language and what it reveals to the researcher when he/she describes the phenomenon through the art of writing and rewriting.

In addition to using the personal journal to aid in decentering and to facilitate openness and transparency, the researcher also used it during data analysis to enhance the accuracy of the description of the phenomena of interest. The researcher had personal experience with Alagille syndrome because her daughter passed away from complications related to the disease. This factor required the researcher to be acutely aware of her assumptions and personal biases when she analyzed the textual data.

The researcher enhanced methodological rigor by using the Lincoln and Guba's (1985) naturalistic perspective to establish the trustworthiness of the qualitative data. The elements of credibility, transferability, dependability, and confirmability formed the basis of trustworthiness of the qualitative data. The researcher addressed these criteria by an extensive audit trail of raw data that included audio-recording of interviews, interview transcripts, field notes and demographic survey results. In addition, the researcher used decentering, kept a personal journal of assumptions and biases, use an adequate sample to achieve data saturation, audio-recorded interviews, and consulted with an experienced qualitative researcher early in the data analysis process to further strengthen trustworthiness and dependability of the data. Lastly, after completion of data analysis, the researcher presented a summary of essential themes, interpretations and findings to two mothers: one mother who experienced chronic sorrow and one mother who did not. The researcher approached these mothers via email and asked them to perform member checking based upon their desire to help the researcher, their interest, and their availability. Their emailed responses verified and supported an adequate representation of their lived experience.

## **Ethical Considerations**

The researcher obtained approval from George Mason University's ORIA prior to starting data collection. She took steps to reduce participant burden and enhance responses by conducting focused interviews that lasted on average approximately 40 minutes. The researcher also reminded participants (who had provided written consent) several times that they could refuse to answer any question(s) and/or withdraw from the study at any point.

The participants in this study were not classified as a vulnerable group population and, thus, further special precautions were not warranted. Each mother in the study selected the time and date that the researcher conducted the interview. The mother also chose a location for the telephone or Skype interview with which she was comfortable. The researcher set in place measures to insure confidentiality and privacy of participants, including securing and coding the digital recording.

The researcher took several steps to maintain participants' privacy. She assigned each participant a pseudonym known only to her and the mother, as discussed previously. The researcher numbered the pseudonym and matched it to the numbered consent. She kept this information in separate locked files to which only the researcher had access. In addition, the demographic information did not ask for names or specific locations of residence (i.e. city/areas). The risk to the participants was minimal because they completed interviews in the privacy and comfort of a place of their choosing and were not be subjected to physical harm, discomfort, or psychological distress. In the event that a participant had reported experiencing psychological distress during the interview, the



researcher was prepared to provide emotional support and to refer the participant to appropriate support resources in their community.

### **Limitations of the Study**

The concern in qualitative research relates to how the researcher interprets the rich amount of data collected from the participants. Streubert and Carpenter (2011) note that the researcher is “an instrument and part of the study” (p.22). Researcher bias, as well as issues regarding trustworthiness and dependability, raises a valid concern that this researcher attempted to address. However, some notable study limitations must be acknowledged despite the selection of the best design and analysis to explore the question.

First, the use of a small purposive sample means the findings will not be generalizable to all mothers of children with rare disease using online health care communication to manage chronic sorrow or other unpleasant feelings. Mothers who did not participate in the study might have had different experiences.

Second, not all of the mothers of children with Alagille syndrome experienced chronic sorrow. The researcher anticipated this possibility beforehand because Eakes (1995) noted that a Black couple in her study involving parents of chronically mentally ill individuals did not report the presence of chronic sorrow.

Third, the data analyzed in this study came from mothers who participated in an audio-recorded Skype or telephone interview. Those participants who chose a telephone interview did not permit the researcher to capture the non-verbal aspects of

communication in humans. Non-verbal communication might have contradicted what the mother actually said or enhanced the significance of her verbal communication.

Fourth, despite vigorous recruitment efforts and snowball sampling, the sample, while geographically diverse, was demographically homogeneous. The experiences of the mothers in the sample might not reflect those of mothers with different educational level, employment status and resources.

Lastly, the researcher conducted a single interview with each participant. The researcher's experiences and knowledge of Alagille syndrome had the potential to impact the mothers' level of comfort and confidence in that the information they shared was being well received and understood. However, it is possible that a second interview may have yielded additional experiences that the mothers may have recalled after the interview concluded.

### **Summary**

The researcher used a phenomenological approach to explore the use of online health communication to manage feeling such as chronic sorrow in mothers of children with a rare disease. This chapter described the underlying philosophy, design, population/ sample, instrumentation, data collection, general data analysis, ethical considerations and the limitations of the study. The qualitative methodology elicited rich descriptive data that will enable nurses and other health care providers to understand and support mothers who seek online health communication resources in caring for their child with a rare disease.

## **CHAPTER 4**

### **Findings**

This chapter presents the findings of the participant interviews. Descriptions of the participants and brief descriptions of their children begin the chapter, followed by thematic analysis of the interviews within the context of van Manen's (1990) guidelines for analysis. The transcribed interviews served as the foundation for identifying essential themes. The researcher conducted numerous holistic readings of each participant's narrative. The readings allowed the researcher to identify significant areas of text that were selected and highlighted, which ultimately resulted in uncovering the meaning of the mothers' experiences with online communication and chronic sorrow.

The conceptual framework for this study was instrumental to informing the development of the interview guide. The middle range theory of chronic sorrow posits that certain events would initiate pervasive psychosocial stress on the mothers; their desire to manage this feeling would lead them to use online communication, which also perhaps could be an identified trigger of the experience of chronic sorrow. The findings of this study, which will be discussed in detail in this chapter, revealed that about half of the mothers reported experiencing chronic sorrow; the other half experienced feelings of fright, anger or being overwhelmed that might be classified as unpleasant feelings.

However, the lived experiences of all the mothers were very similar with regard to how they used online communication to manage their various feelings.

### **Description of the Participants**

A purposive sample of sixteen mothers of a child with a diagnosis of Alagille syndrome participated in this phenomenological study. The mothers ranged from 31-53 years of age, with a mean age of 38.94 years. Participants were predominately Caucasian/White (81%; N=13), with the remainder being Black (19%; N=3). The majority of the participants were college educated (88%; N=14), married (75%; N=12), and employed outside the home (88%; N=14). Fifteen participants resided across the United States, while one participant was from a European country. The majority of mothers stated at some point during the interview that they had other children in addition to their child with Alagille syndrome.

The children who were diagnosed with Alagille syndrome ranged in age from 6 months to 17 years with a mean age of 7.5 years. The average age of the children at the time of the Alagille syndrome diagnosed was 11.5 months, with the median age being 2.5 months (the mean is much higher because one child was formally diagnosed at 12 years of age). The children had a variety of additional medical diagnoses associated with Alagille syndrome, with the predominant diagnoses being peripheral pulmonary stenosis and congenital heart disease. Five children had additional medical diagnoses not generally associated with Alagille syndrome. These diagnoses included Legg-Calve-Perthes disease, celiac disease, hemiparesis, vitiligo, attention deficit hyperactivity disorder, and food allergies. Five children were liver transplant recipients. See Table 2

for a brief demographic description of mother’s age range, highest level of education and the age of her child with Alagille syndrome.

Table 2. Demographic Data of Participants

<b>Participant</b>	<b>Age</b>	<b>Mother’s Highest Level of Education</b>	<b>Age of Child with Alagille</b>
Ann	50-55	College	17 years
Beth	30-35	College	19 months
Clare	40-45	College	10 years
Dawn	30-35	College	9 years
Elle	30-35	College	2 years
Faith	40-45	College	11 years
Gail	40-45	High School	8 years
Hope	40-45	College	4 years
Ivy	30-35	Graduate School	2 years
Jane	45-50	College	13 years
Kate	30-35	College	3 years
Lee	30-35	College	14 years
May	45-50	College	14 years
Nan	40-45	Graduate School	11 years
Olive	30-35	High School	6 months
Pearl	30-35	College	4 years

*Note.* Participant names are pseudonyms.

### **Chronic Sorrow and Unpleasant Feelings**

Interviews revealed a rough start for the mothers of children with Alagille syndrome, especially for those mothers whose children were diagnosed when online communication was in early development. The overarching theme that emerged was online communication proved essential to the Alagille syndrome community in the management of chronic sorrow and unpleasant feelings. However, described events that triggered a mother’s unpleasant feelings or chronic sorrow included online

communication encounters, in addition to developmental milestones and other anticipated triggers.

The interview process began with the mothers being asked to recall the feelings they experienced when they were first told their child was diagnosed with Alagille syndrome. The mothers described many feelings, including anger, fear, helplessness, overwhelmed, guilt, shock, worry, and sadness. These feelings were reported as periodic and recurrent since the initial experience at the time of their child's diagnosis. The mothers who provided evidence of chronic sorrow reported permanent, periodic, pervasive feelings of sadness that were consistent with the theoretical and operational definition. Olive, the mother of an infant and early in her journey with Alagille syndrome, illustrates the chronic sorrow she experienced as her infant daughter had a medical device that impeded her abilities:

I see other parents with kids that are normal or I have my girlfriend [who] has kids, we were pregnant at the same time. Her baby is huge and happy and they are doing all this normal baby stuff and that makes me sad that I can't do all that stuff yet.

Pearl, the mother of a preschool aged child provides evidence in her response of the pervasive nature of the chronic sorrow:

If I allow myself, I'll get sad about it [the situation of having a child with a rare chronic illness] because I see those kids playing, I see my beautiful girl with these tiny, tiny, arms (makes a circle with her fingers to indicate size) and tiny, tiny legs and the jaundice.

Lee's (mother of a teenager) excerpt highlights permanent and periodic nature of chronic sorrow and discusses triggers that bring up the recurrent nature of the sorrow:

She [her daughter] is the size of a 5 year old, so some kids think she's a dwarf or a midget or things like that, but anyway, she can't take driver's ed, she hasn't started her menstrual cycle yet, while her friends have. So because we're hitting milestones; like every time we hit a milestone I kinda cry sometimes in the bathroom. I'll be strong for her because then she'll ask me certain questions and I'll try and make the best of it; then once I get by myself I cry.

Mothers experiencing feelings of chronic sorrow appeared to have strong, solid and consistent support from three types of people: 1) the online community, 2) family/friends, and 3) medical/health care providers. In contrast, mothers who experienced other unpleasant feelings appeared to have one of the three significant supports impaired in some way. Dawn, a single mother of a school age child described her initial and recurring feelings of fear. When her child was first diagnosed, Dawn was with her spouse, but she did not experience the support she needed, "[h]e was really receptive as far as listening to the fears, but he wasn't a good responder and in another way left me not only feeling fearful, but very helpless and alone". After Dawn and her spouse separated, while Dawn had some family close by she was left to manage her feelings of fear. Dawn describes an incident in which highlights the permanent and pervasive nature of her feelings of fear:

I remember one time; Ann [child's pseudonym] had a couple of burpies you know, receiving blankets. They were covered in blood; it's like a bloody

massacre. I took them to [the child's doctor] and I cried in his office. I said, 'Nobody understands, nobody understands. I don't know what to do. She was up all night digging. I've got blood everywhere. I think I'm crazy'. You know and I'm like "how can I help her, what can I do?" He let me cry in his office and he told me it was gonna be okay" and, and I was thankful for that.

Lee was a single teenage mother when she gave birth to her child with Alagille syndrome (now a teenager). She was the only mother to express anger as an initial feeling, "initially I was angry; I didn't understand why because I felt that I had done everything that I was supposed to do in order to have a healthy baby". This anger became periodic and pervasive and is highlighted in these following excerpts:

I was angry at the doctor as well because they had tested me for Down syndrome, you know [and] the typically things they test women for when they're pregnant. I just could not understand for the life of me how come they had not tested me for Alagille syndrome. It was not until I went to genetic testing, realized that it was rare and sometimes with a lot of those rare diseases, your OB/GYN doesn't test for those things; they just test for the basic stuff.

While, Lee readily identified her mother as her biggest source of constant support, she did not have online support early in her journey nor support from others who in a similar situation. However, in Lee's case, the feelings of anger were not permanent as she slowly developed strong support networks in online communities, with other mothers of children with rare diseases and solid relationships with medical professionals. In addition, she has



amassed the experience of parenting a child with a rare chronic illness. Lee described how she has reflected on her experiences and works helping others:

I feel grateful because (voice cracks) I didn't see it then, but I see it now. I've been able to connect with a young lady that I work with and she started an organization called [names organization]; her daughter has a rare disorder and as a result of her own frustrations she wanted to help children and their families who suffer from rare disorders with emotional support and because I am part of that, I've really been able to talk to other moms who really don't quite understand.

They [mothers] say 'Why me?' Then I'm able to tell them my story because I was a single mom when I was pregnant with Nana [her daughter] and I was so hostile.

Lee states, "Now at times I feel more sad." Lee's evolution of her feelings best supports how impairment in some component of support may inhibit the experience of chronic sorrow.

Kate, the mother of a preschooler is married and is surrounded by supportive family and friends like many of the participants in the study. However, she remains fearful as she resides in an area where access to specialists and those knowledgeable about Alagille syndrome appears to be challenging, so she experienced an impairment in medical/health care providers. She voiced what many mothers shared in their narratives, "everyday is just a worry for me because I just don't know what every day, what each day is gonna bring."

While these mothers differed in terms of their emotional response to having a child with Alagille syndrome, the online communication use and experiences was

essential and consistent among all the mothers. Therefore, thematic analysis conducted in this study describes recurrent themes and commonalities among all the participants in their use of online communications to manage chronic sorrow and unpleasant feelings.

### **Thematic Analysis**

The researcher identified four themes related to online communication: 1) connectedness, 2) online triggers, 3) empowerment, and 4) seasons of online use. These essential and interrelated themes provide a more complete description of the lived experience of these mothers. The essential themes were developed from several subthemes, which are listed in Table 3. The identification and relevancy of the thematic elements contributes to a rich understanding of the experiences of these mothers and hopefully begins to broaden the discussion of experiences among all women of children with rare chronic diseases.

Table 3. Themes and Sub-Themes

<b>Theme</b>	<b>Sub-Themes</b>
Connectedness	There is hope Being part of the community
Online Triggers	We could lose our child Overload via social media Medical piece
Empowerment	Deciding the best place for information We're the experts
Seasons of Online Communication	Using online communication more Using online communication less

## **Online Communication is Essential to a Rare Disease Community**

Mothers expressed their feeling that having a child with a rare disease can be a very scary and lonely experience. The mothers had to adjust to a new way of building relationships and connections, in addition to coping with their child's illness. While the mothers shared their joy at being able to meet others via online and establishing a "personal connection", most mothers shared their preferred interaction was face-to-face meetings with other parents of children with Alagille syndrome. Kate shared the experience of many of the mothers when she said,

I wish that there was somebody closer that I could get together and talk and kinda just share our experiences that way, but I am very fortunate for the online part of it. But I kinda wish that there were somebody that I could speak to face to face.

Kate's further described the ideal personal connection involved meeting,

[S]omebody that I could meet and talk with maybe once a month; maybe get our children together, so they can meet and play. I [would like to] just form some kind of a friendship and a bond with someone who is going through the same thing that I am.

Pearl also captured what several other mothers experienced with regard to stronger bonds formed in face to face meetings. She discussed how the online communication was wonderful for the initial connection, but the face to face meetings were ultimately the best form of communication. Pearl tells of her experience of how the mothers of children with Alagille and some other rare liver diseases met online and then were able to organize a face-to-face meeting at a park, "We brought our medication to compare

(laughs). We could talk about all that stuff we talk about online and then now in person and bonds were forming.”

The overarching theme that emerged from the participants’ narratives indicated it was essential to use online communication to: 1) access information, 2) increase knowledge and empowerment, 3) help in advocacy for their child, and 4) to establish relationships with others who are going through a similar experience. Participants reported their experiences were overwhelming, with little information or support from health care providers early in the diagnosis phase. Mothers and their families were left to try to make sense of the rare diagnosis that was given to their child. The journeys for these mothers began in a similar fashion, with a general query using a popular search engine such as Google or Yahoo.

Mothers of older children described the process of trying to find information, resources, and support online. The early online communication years were fraught with limited and dated information/research. For example, Dawn, the mother of a 9 year old, reports:

Every time I looked up a symptom of Alagille or looked up Alagille in general at that time I could maybe get three to four small documents or pages that had anything that had to do with Alagille...I couldn’t pull much up at all, but when I did read it was very scary; I really didn’t know how to feel about that.

In addition, mothers using online communication in the early years reported the predominant online formats involving asynchronous communication, which could be frustrating, difficult to use, and involved waiting for responses longer than could be

afforded. Pearl, the mother of a younger child, who used to the Alagille Alliance message board first to connect with other parents states, “I wrote a message to introduce myself and people reacted on that, but it was really on its way out because it was a very slow board, [there were] few reactions to what you were posting”. Ann clearly states, “the bulletin board is a little more complicated to get into.” This sentiment was further echoed in Gail’s comments, “I don’t use the board anymore because nobody else uses the board. I think the board was too hard to navigate.” May used email but, “back then I guess I probably really wasn’t really aware of all the different resources that were offered.” However, despite these challenges, the mothers reported that the ability to connect with someone who had a child with the same disease was invaluable and essential to their ability to provide comfort and support to their child. These interactions also gave them knowledge to share with others, including health care providers. Nan, the mother of an 11 year old sums it up, “I think the personal connection with other parents and the personal experiences, that’s what gets you through”.

Over time as online communication evolved, mothers of older and younger children described the wealth of information available as well as the ability to enjoy immediate responses via online communication formats like social media. Gail states, “there is a lot more information out there than when I was Googling.” All the mothers reported using Facebook to communicate with other parents of children with Alagille syndrome. The ease of the mothers to connect to others using Facebook was described as “it’s just right there when you’re on Facebook; it’s just so easy to get that notification and you just go right to that group” and “Facebook is an easy app that’s on a lot of phones

and you can just check it and it's right there." Olive, a new parent to the Alagille community, states:

Facebook has been nice. I've been able to talk to some moms that have similar problems and through Facebook I'm talking to the Alagille parents. Every kid is different; they all have different issues, but it's definitely nice—to talk and kind of see what her future's gonna be like; the moms that have older kids and they're doing well and all that stuff.

However, the dramatic increase in the amount of information available online has led the participants to describe new challenges which include trying to decide the best places to get information. Hope describes the activity new parents generally engage in when seeking information on Alagille:

If you are a new parent and you just go and Google it you will get all sorts of things and old studies that will tell you that your kid will definitely have the mental disabilities and all that; you know like the old studies. In terms of getting the consistent new information; as a new parent is very hard.

Dawn further describes how information is more available, but the struggles of what does all this information mean and how it impacted her emotionally:

My negative for the internet is not when it comes to communicating with other parents or people; it's the information that I read. Early on Wikipedia had what Alagille was and it scared the Dickens out of me. I think, um, Web MD has some information on Alagille and I printed it off and showed it to my family members and we just sat around and cried. My daughter's gonna die you know, she's not

gonna make it. What am I gonna do? The information I was getting was very doomsday; it was a doomsday scenario. It was very scary.

The advantages of social media sites like Facebook also bring a down side. The mothers report consequences in using this online communication tool includes encountering more of the lives of families. Gail discussed some of her observations regarding her Facebook use experience:

You know on Facebook you don't just all of a sudden hear all about Alagille. You mostly hear about their life; just different lifestyles. You know you begin to meet -- see people that you would never even probably be friends with and then all of a sudden you are friends just because the Alagille. So you know there are a lot of times I have to like block people. I've never had any negative talks back and forth and stuff. So you know it's stuff that kind goes against your own moral code that you see.

Lee discussed several online communication formats. She describes an experience with a message board that was negative and offered a reason why Facebook for her has been a more positive experience and how encountering the lives of families online could offer a protective factor:

I kinda stay clear of certain areas particularly [names a message board]. I don't know if it is like that anymore, but there was a time where the chat boards turned into people being mean and cruel to other moms. So, ever since that happened I really just tried to stay clear [of that board]. I should give it another chance, but I

haven't because I believe in there is power in the tongue and I just sort of want to keep my circle positive.

I do connect with people through Facebook, personal email and things like that. I am really at a point right now where I pick and choose who I want to deal with because I HAVE to stay positive when it comes to dealing with a child with a rare disorder. So, I choose not to deal with certain people. I think with Facebook has been pretty positive overall. One reason why I think that is because when you're on Facebook, not only do you have the people with Alagille, but you also have people's friends and families. I do think that, people are less prone to come to your page and say something negative or derogatory especially when you have other people who can defend you.

Social media site encounters often include exposure to families dealing with the deaths of children with Alagille. The mothers reported deaths definitely posed a challenge in terms of the need to be connected to other families and supported in the Alagille community, while encountering child loss related to Alagille. Gail described how one develops closeness through frequent online communication and sharing of the experience of such a rare condition. Gail states, "when you see something happening to the kids, I mean the Alagille family feels like your own. They feel like family to me so it is really difficult for me to see these kids struggling". May who belongs to several Facebook groups related to childhood liver disease including groups dedicated solely to Alagille states:



I had to leave the Facebook liver group a few months ago because every time a baby died and I read about it, I would be a wreck for 2 or 3 days and I just finally had to leave the group—I just, I just can't do this anymore. I feel so bad for the parents and then just you know. I just had to leave because it was getting to be too for me.

Faith and her teenage daughter frequently visit Facebook and Alagille pages. Faith shares her pain and challenge of needing to participate in the online communication, but trying to balance reality with optimism of her daughter's future due to reading about children loss related to the complications of Alagille syndrome:

Just when we see kids with the same disease that, it's just really hard, even though we don't know them (voice cracks a bit). It kinda hurts a little (sounds teary).  
Sorry. I know you just went through that [death of a child]. That can really get you to going. This is the reality and last year seemed to be a lot [deaths]. I don't know if it's because more people are on Facebook. In the beginning we didn't hear that as much. Charity [pseudonym for her daughter] this is the reality you know. She's almost 12 so it's just that and leading her through that and it can be kinda hard.

Hope summed up the sentiments of many of the mothers. She recommends "I think that everybody just needs to find their comfort. I think that burying yourself in the bulletin board and Facebook and constantly being there can also really get you down".

### **Theme One: Connectedness**

Online communication is dynamic, fluid and evolves in response to parents needs and technology advances. Mothers viewed their ability to become connected to an online community for support as critical, “it felt like a lifeline for a lot of us”. The isolation and lonely feelings of being a mother of a chronically ill child are particularly predominant in the rare disease community. The researcher noted comments like, “I think community is incredibly important to know that other people are going through similar things.” “You need to communicate because it’s hard when there is not a lot of it and nobody knows what it is.” “There’s nobody – I mean it’s such a rare disease there is nobody local, nobody around me, at least ever heard of it, you know. They are always like ‘what the heck is that?’ ” “I think the personal connection with other parents and the personal experiences that’s what gets you through. That [online communication] was just key in me feeling connected and not being alone in it.”

The ability to feel a connectedness with other moms when one is dealing with a rare disease is well illustrated in a response by Jane. She notes that the connectedness moms can derive by communicating online gives rise to some sort of solidarity. In addition, Jane’s excerpt highlights the concept of upward and downward social comparison, which is often a component in chronic illnesses:

How your child always looks a little different than everyone else’s child or they don’t progress at the same rate and I guess that sort of solidarity is really important because it make it feel like you’re not, not something so terribly wrong with you child. When you look at them compared to other Alagille children,

they're doing fine, they're doing just what Alagille kids do instead of looking at the world at large and other's kids and how they progress.

The internet has changed the level and frequency of communication among mothers of children with Alagille syndrome. All mothers reported they used the internet in some capacity. The internet has allowed mothers to communicate, seek support and information, and form a sense of connectedness with other mothers from all over the world. As internet communication has evolved, mothers have evolved with it. Currently social media, specifically Facebook and the search engine Google, are reported to be used most frequently to get a sense of feeling connected as well as obtaining information regarding Alagille syndrome.

Nan described that when her son was diagnosed 11 years ago she used the Alagille Alliance message boards frequently. She discussed a unique feature that is not typical of social media platforms and how this was helpful to her feeling connected.

I used the message boards a LOT, a lot. You know and the thing with Alagille is you have multiple organ issues you know and, and some people can relate to the lungs, some people can relate to the kidneys issue and that part really helped because you could really connect with the people who had the same part of the syndrome that your child had.

Lee started her journey with the Alagille Alliance message boards 14 years ago. She made a transition to Facebook and initiates contacts with many moms through private messages and emails. She discussed the internet globally in terms of what it has meant to her in terms of connectedness to other mothers dealing with Alagille syndrome.

I just feel like the internet give you more of a personal touch with individuals and you can always chat with them. You know, um, “hey how’s your kid?” and they ask you and then sometimes people post what they’re going through and you can make comments and things of that sort. So, I think the internet has been a wealth of resources and knowledge and connections for me.

Hope’s experiences as a mom of a 4 year old child reflect the connectedness and ability to relate to a larger number of individuals. She started with the Alagille Alliance message boards when her daughter was first diagnosed at 3 months. After a time, she desired to become connected with a larger network and once again, Facebook fulfilled the need.

Eventually they [Alagille Alliance] started the Facebook as well. That’s the place where you kinda see people and you can see people are doing and how their kids are doing. So, I see a lot of that right now. Initially, it was just the bulletin board and uh, there just seeing people communicating there, it seems like a very small community you see maybe 3 or 4 people posting on there on a regular basis.

Dawn, like Lee, is the mom of an older child who started out using the Alagille Alliance bulletin/message boards to experience the connectedness that is so vital to managing a myriad of feelings. The feelings of connectedness that came from being a part of an online community helped to validate her feelings and abilities as a mother of a very sick child.

So I really depended on that board and I would read other parent's questions and through that board it really helped me know that my feelings of confusion or am I being a good mother, is all this blood on her sheets normal? It really helped me; it grounded me a little bit to know that I wasn't crazy and that the feelings were normal.

Dawn then goes on to describe how she establishes connectedness using Facebook. She gives very compelling reasons as to why this format works for her in feeling connected to other moms.

I solely rely on Facebook because I can reach out to anybody. I can leave them a message. I can get phone numbers and talk to them. I learn people's stories without them knowing. I watch their pulse and learn about their children without having to intrude in their lives and if I do want to say something I can pop in and say something and introduce myself. If they want to reach out or connect with me, I am more than willing. [Also] Facebook is where you can keep these people in a contact list and reach out to these people when you need them. I like that, that's how I do it.

**There is hope.** A recurrent sub theme essential to connectedness in the mothers' narratives was that being connected allowed them to experience hope. The mothers shared the ability to see pictures and posts of children who were doing well (especially older children and adults) provided optimism and a reason to be hopeful. Nan states "the Alagille Alliance that website and hearing other mothers' stories and, and the grown-ups

who have Alagille syndrome and just personal experiences for ME, it was the best". Jane, mother of a 13 year old daughter, discusses how she was able to share online communication information with another mother about her daughter; this gave the other mom hope and reinforced her [Jane] hopes for her daughter,

It was really helpful for her to see that her child could grow up or her child could get bigger or get better or feel better you know, there is still hope where you do come through this really, really tough period.

Olive reports "Just kind of talking to these people [Alagille parents] is definitely making life better. It's definitely making me look more positive toward the outlook".

Beth, the mother of a toddler, described a difficult beginning when her child was diagnosed. She used online communication to find information about the disease and through online support was able to find hope as well:

In the beginning, I would say it was bad because when you first research something that not much is known about it, all you really see are the negative aspects you know; basically the morbid parts. Then as I got more in depth my researching it and stuff like that and speaking with other people you know either through chat or email or whatever else, it became somewhat more positive. You see some of the success rates or you know the kids living longer than what they are expected to or I should say what the doctors expect them to really.

**Being part of the community.** The importance of being part of a community that actually understands a rare chronic illness was a very important recurrent subtheme. The mothers could pose questions and express their fears and frustrations without being judged, offering a safe place for many to express positive and negative feelings they experience. Kate the mother of a preschool child described how she benefitted from being part of an online community. She reports:

I think it's great because you can pose any question, you should be able to feel comfortable because everybody that's on there is going through the same thing, Alagille. So I just think being able to pose any questions that you may have and then having others respond to you is very helpful.

Many of these mothers had other children, so they could get suggestions and emotional support as to how to involve and reassure their children who did not have Alagille syndrome in the care and support of their sibling with Alagille syndrome. Lee's excerpt illustrates how she suggests to moms in the community that they do not forget the siblings of the child with Alagille:

I also feel that is very important not to forget the other siblings. Even if it means that you give that child [the child with Alagille] a brief talk in his room bring the siblings on. You know, just stuff that simple because my [other child] she said 'momma, why everything always gotta be about Nana?' and it just hit me and I was like wow. When I stepped back from it I didn't realize that people was always like 'How's Nana?' or I always talked about her.

They also felt they were providing a foundation for their child to be able to participate and communicate with others who had the same disease. Hope discussed how being part of the community is important and will become more of a factor as her daughter grows and develops:

I think that you know, I think it's important to be aware of it and I think it is important to be a part of the community and this is the children that we will see for years to come and I think it's important for us and for Maria for that matter. You know, I think when she as she gets older she will want to see other kids with the same condition. So, it's important for us to be a part of the community.

Faith, the mother of an older child, was not that proficient with the internet and did not use it much prior to her daughter's diagnosis. After the diagnosis, she describes the invaluable connection she experiences when she begins to interact with parent online:

We had internet, but I had never really emailed and all that stuff and searched the web. So, I was really clinging on to them as parents knowing what I was going through. They were really helpful to have that connection.

Mothers of older children "veteran" moms of Alagille were able to transition to different roles in the community. Lee now sees her role in the community as that of a "problem solver" and encourager:

I play the role of encouraging and so I share different experiences with people. I always come across as the problem solver. You know when there's a mom if



she's tried certain medications she may ask if Nana [her daughter] has ever taken them before. For instance Rifampin, I tell the mom's to make sure they wipe the children's mouths out real good because it can cause discoloration; just different little things that I've been through. I can like tell other moms. So, I play more of the problem solver I believe when it comes to the moms.

The online community is very important to all the mothers, but several mothers did note there were some downsides to online communication, like social media. As cited earlier, the mothers described being exposed to the lives of their Facebook friends, which included lives and communities outside of Alagille syndrome. However, the need for community is so great the mothers are willing to stay connected and continue in the community and just employ tactics that would offer a buffer or protective feature. For some mothers, sharing some of their deeper feelings or experience in an online community format such Facebook is not desirable as evidenced by Clare's narrative:

I think community is incredibly important; to know that other people are going through similar things. It's a little hard on Facebook because you are sharing so much of yourself with the community [and] not just the Alagille piece. You're sharing everything. I don't post very often on Facebook and that's part of it. I don't want to necessarily share everything about myself (laughs). I want to say that probably 75% of my friends on Facebook are from the Alagille community and the remainders are family and friends. That is primarily that is how I use Facebook to just stay connected. You're learning about all these other things that

you don't (laughs) need to know about people. I wish there was a better way for the community to share their experiences because I think that is really helpful because you know it's hard. I didn't feel comfortable sharing some of the stuff with the community because I may be met these people once at a symposium; I don't necessarily want to share those thoughts.

Belonging to a community also helped the mothers to obtain tips for managing symptoms unique to Alagille, as well as helping the mothers put their experience within a context. Ivy voiced the experiences of several mothers in the following excerpt, which describes a supportive approach to the management of itching that is a frequently discussed topic in the community. In addition, the excerpt highlights how Ivy was able to see her daughter's itching and scratching in the spectrum of Alagille syndrome. Ivy states:

I've gotten some amazing suggestions too. We are now using Velcro rollers and I don't remember who put that on there, but Velcro hair rollers they [children with Alagille] can get some relief from itching, but it doesn't destroy the skin. That was like the best tip of all time. So just connecting with other families to hear what's working for them.

Alagille.org has been super helpful; the parent forum there just connecting, hearing what other parents are doing helped. I was just thinking about how it's put her itching in perspective. I think it's really bad where she is up all night, but then I read about kids that really scratch themselves raw. Well, she's not that bad so

that puts a little bit in perspective as we are thinking about our next steps in her treatment.

In some cases, being a part of an online community has translated into being able to meet in person, which most mothers agree is the optimal interaction. Pearl discussed how a support group she started online was able to coordinate a meeting of several mothers of children with Alagille syndrome:

We brought our medication to compare (laughs). We could talk about all that stuff we talk about online and then now in person; bonds were forming and that is [what] I think is the great thing about getting people who are going through the same things, or have gone through the same things together because, a doctor knows a lot about it, a nurse can comfort you and talk more about it, a social worker can, calm you down, but there's NOTHING compared to first hand information from people who have gone through the same thing.

### **Theme Two: Online Triggers**

Online communication has the ability to trigger chronic sorrow and a variety of unpleasant feelings. The researcher identified the common triggers of too much information (especially regarding early deaths), information overload regardless of the online format, and/or the “medical piece”. Mothers seeking information as well as mothers heavily involved in social media all experience the ability of this powerful communication form to trigger unpleasant feelings.

**We could lose our child.** This subtheme was by far the most prevalent and powerful trigger for the mothers. All mothers experienced unpleasant feelings when encountering parents' posts and videos online of their children who died from complications of Alagille syndrome. The mothers' observations of child loss online reminded them of the fragility of their own children and the highly variable and unpredictability of this disease. Lee's excerpt illustrates the unpredictability of the disease course. Lee describes a child and her family she followed on Facebook. According to Lee's observations and interactions with the mother, the four year old girl appeared to be doing well, but then the child died suddenly:

Anytime we lose an Alagille child I cry. Last year we lost one and I cried. I was at work and I actually had to leave work it bothered me so bad. For one, I had no idea that anything was going on and so it just, just made me cry because it was just like sudden and why.

An early death is a realistic outcome of Alagille syndrome that the mothers have to acknowledge can occur. In addition, a sudden death is more likely to occur for many of these children.

The majority of mothers avoided reading blogs on Alagille syndrome as they perceived them as containing negative information about children who were generally in a poor state of health. Elle described her experience, one that was shared by most mothers,

I try really not to [view blogs] that much because sometimes they sound so dire. I kinda don't want put myself in that kind of mind set like 'oh this is a possibility'.

Alagille as you know it affects every child differently.

The mothers mostly learned of deaths of other Alagille children on Facebook posts and You Tube, when seeking other types of information or support. Pearl wonders sometimes why she subjects herself to Facebook groups related to Alagille, as she wonder how the parents are feeling:

Sometimes a child will die and I'm in tears every time. Every time a little angel dies I'm distraught because I think of the parents and what they must be feeling.

I've been, wondering if I should be on that Facebook group at all. Every time that [a child's death] happened I wonder what I am doing here?

Gail likes to watch videos of the children with Alagille and imagine life as her school age child gets older. In her quest to catch a glimpse of the future, Gail is forced to encounter a harsher reality:

You Tube, You Tube and don't You Tube Alagille, then you see the kids that have passed away and then it makes you sad. I've done that. You wanna see what kind of future they're gonna have and you see all the sad stuff too; that's hard.

Ivy experiences unpleasant feelings from online communication. Ivy identified feeling of sadness and acknowledges the encounters were hard to experience. However, she also identifies feelings of guilt for having a child who is not on the severe end of the Alagille disease spectrum:

Anytime there is sort of a memorial or we are talking about kids who have lost their battle that is a little bit hard for me in two ways. First of all, it brings to reality; it makes it even more real what is a possibility for her. Then there is this other piece where I feel a little bit guilty that because here I am really worrying about my kid who's in this amazing place where other parents are not in that place and that's hard for me too. You know, I feel a little guilt of trying to access, I don't know what I am trying to say here but I feel I am taking resources for some parents who probably need it a lot more than I do

**Overload via social media.** This subtheme like the other two subthemes, *we could lose our child* and the *medical piece*, addresses the amount of online communication can trigger the unpleasant feelings experienced by the mothers. Ann's excerpt describes what several mothers expressed. The mothers have so many Facebook "friends" that they know in the context of Alagille, but do not really know as people.

I'm friends with a lot of these people on Facebook some of that is just a product of Facebook. I know them because I am friends with them on Facebook. They post tons of stuff that is of no pertinence to me and so then that gets like almost overload for me. But, I don't want to defriend them because I know them; they have a child with Alagille syndrome and you know somewhere you can support each other too. So "like" to all these other people, sometimes it gets overwhelming. It's like I know too many people. I know of them, but I don't really KNOW them.

The rarity of the disease trumps the mother feelings sense of feeling overwhelmed, so Ann, like many of the mothers feel a sense of duty and obligation to remain “friends”. This sentiment was further supported in an earlier comment by Gail. Gail described how she was “friends” with people she would never be friends with if it were not for the commonality of having a child diagnosed with Alagille syndrome. She elaborated further that she was exposed to behaviors and people that went against her own “moral code”. These encounters within the context of social media along with the frequency and ease of access to other mothers created an environment where the mother experienced feelings of being overwhelmed and overloaded with communication. However, despite the risk of encountering unpleasant feelings from too much of this form of online communication, the mothers know the importance and value of support when dealing with a rare disease like Alagille syndrome and go to great lengths to support the community.

**Medical piece.** The medical piece subtheme was described by many mothers as an online trigger for unpleasant feelings. There were many occasions when the mothers were searching for information regarding Alagille syndrome in general or on a specific procedure. Many mothers reported some of the information they found, while helpful or even critical, could also trigger unpleasant feelings. Kate reports, “I did read this one article and I remember reading about the survival rate age and that really kinda scared me.”

The amount of information now available online about the medical interventions, treatments and procedures had several mothers questioning if they were making the best decision for their child. Clare describes how this occurs:

Well I think there are 2 different types of online communication. There's the medical piece where you're searching through medical journals or articles or whatever trying to make sense of it from that stand point. You know that can be I don't wanna say almost dangerous it, almost makes you spiral a little bit because you start thinking 'oh well I need to have this test done or I need to have that test done or oh I wonder.... You start reading into little things.

Some mothers with strong family support and the ability to rely on others to gather information minimized their exposure to online triggers especially when the diagnosis of their child was fairly new. Faith was able to rely on her husband to sift through information and only expose her to the helpful pieces that she needed to manage her daughter's care.

My husband actually did the research. He wouldn't let me look things up online because it was, there was so much information and a lot of it was bad (laughs). So he was really helpful while I am dealing with a sick one. He's kinda doing the research stuff and handing me the pieces of information that I needed.



### **Theme Three: Empowerment**

The mothers recognized the value and importance of knowledge and information in their life with statements such as “online sources for making yourself literate—it’s a must, it’s a must; if you’re gonna advocate for your child any way” and “I wanted information.” The mothers agreed that it is best to get information from a variety of online sources. The mothers’ ability to feel empowered due to the knowledge and information they acquired allowed them to help their children and provided a sense of relief and some level of control over the situation. The use of various online communication formats provided the emotional support, immediate support and empathy needed to develop the feeling of being empowered and capable to advocate on behalf of their child.

**Deciding the best place for information.** Many mothers mentioned that due to the rarity of Alagille and its highly variable presentation in each child they wrestled with the best places to get information. “It’s hard to tell who’s really the go to people; where really is the best place to engage. So I think that was a little confusing at first in using knowing the best places to access”. Oftentimes, the health care providers were not able to provide them with much information because “we had a hard time getting information from doctors” or if information was shared it was not necessarily helpful or accurate. Clare shares an early encounter with a physician that illustrates this point:

He [the physician] was very technical and he was also very certain about the path of Alagilles and what it entails. He [her physician] was like he [her son] was

going to have a feeding tube, and he was going to have this issue and he is going to have that issue. You know the path of Alagille is different for each child. He [the physician] was kinda setting up those expectations for us and that WASN'T necessarily our path.

Hope, like many of the mothers in the study, would like to be able to get information and help from health care providers and professionals. However, sometimes having a “hard time getting information from doctors” includes the health care provider withholding information. When information is withheld from mothers, this can complicate their online search for the best information to aid them with knowledge of the disease and ability to make decisions. In addition, mothers have the potential to become frustrated because of the lack of guidance or assistance by these professionals to whom the child's health is entrusted. Hope states:

I would say to me the worst feeling is when I have been managed by a doctor or a nurse; where they decide that they are going to give me a limited amount of information instead of all the information. I think from the medical community, for me that's the worst possible feeling. We can take it, we can understand it. If we can't understand it, we can research it; you can explain it to us. I think information is important.

Online communication allowed the mothers quick and easy access to current information and research. However, the mothers wanted help in understanding and adapting this highly technical medical information and jargon they found online. Ivy the

mother of a toddler illustrates how mothers of younger children are finding much more information and have to do so without the benefit or guidance from health care providers:

There are a lot of different places that have information, some with more detail than others; some with better information than others as far as with that detail piece. I don't think there was a lot of guidance necessarily from medical staff. The [Alagille Syndrome] Alliance is really one of the best places to get the information; that's where the people are, that's where the attractions, that's where I could find the answers to my really specific questions.

The mothers in this study out of necessity became quite adept at locating resources online and using online communication. Nan, the first mother to mention the C.L.A.S.S. (Children's Liver Association for Support Services) website (she was the 14<sup>th</sup> interviewee), explains in this excerpt why this site was so useful for her when her son (now 11 years) was first diagnosed with Alagille:

It was, first it separates the different genetic issues you know, biliary atresia was one. It had them [childhood liver diseases] all on there; it was kid focused; so it was just kids. When you first start looking at things on the internet, it brings all the adult things in too and of course you don't want that. It [the website] was done in very simple terms. Each one of the definitions of what would happen, the symptoms, what was going on with each one of the liver diseases; it was in layman's terms, very much so. Then they spotlighted kids there were other families that you could connect with there as well; very simple and written in

simple terms and towards families. It talked about insurance and how to deal with getting insurance to pay for certain things; I has some of those issues before. So, that's why I like that one.

Pearl elaborates further on her search strategies to get the most information she can about Alagille to care for her preschool aged daughter. As she reports she learned to search by symptoms and conditions associated with the disease:

I didn't only look up what was stated about Alagille, but I took a broader view. [I] looked up articles about failure to thrive and different kinds of situations [like] malnutrition. I could see in Cara when I needed to call a doctor, or dietician or anyone to help about the itching. I looked up what was written about adults and itching and what to do about that. Then I found a website called Scratch Sleeves, it's a sleeves thing. She [her daughter] was scratching all over her face at night and so I ordered those. They are for kids with eczema, but those are itching too. So I went online and didn't only try to get information about Alagille, but all of the symptoms and the growth.

In addition to general medical information and knowledge, the mothers wanted also wanted practical advice and with regard to the unique needs of their child and situation. The mothers appreciated information validated by personal experiences shared online. A good example is related by Pearl who expressed her need to learn about how to manage the intense itching experienced by children who have Alagille syndrome. Pearl reports,

I was very much focused on the itching part of Alagille, Cara was experiencing. So I read every post there was about itching. It was a positive addition to my search for information and things I could do to help relieve itching and what not with my daughter.

As cited earlier, Ivy mentioned how she got “the best tip of all time” by consulting the online community about managing itching in her daughter and was advised to try Velcro rollers as they provided relief without damaging the skin. This was information shared by “veteran” parents who understood exactly the mother’s need to be able to offer her child some level of relief from the extreme itching.

The mothers ultimately were able to decide the best places for information by personal experiences involving observations, health care providers, and other parents of children with Alagille syndrome. Hope provides a very thorough response which describes the approach of several of the mothers when trying to determine the best places to go online for information:

So the bulletin board [Alagille Alliance] was, considering it was such a small community, it was good place to ask questions and things that were kind of common in Alagille... you know it’s [Alagille Alliance board] so teeny and you take it as such; it happened to another kid or maybe two other kids and so I wouldn’t take it as the proper medical answer; if I need medical answers than I ask them of the medical community. Facebook to me is not the place where you share to kind of get medical information, it is more of a place, and well it’s a

community. You see what is going on with other people; you stay in touch with them so, I treat it as such. Once again, it's not medical; once again it's not a place for medical information. I see a lot of confusing medical information [on Facebook]. Somebody will ask should I use Motrin or Tylenol and you get all sorts of answers. So, it's not a good place to be asking or getting medical information from what I can see. I constantly see [Facebook posts] 'Should I not be giving my kid Tylenol or Motrin?' And you look at the responses and you probably see the same thing, full of responses people say 'No my doctor says to never give Motrin' and then another post says 'No, my doctor says to never give Tylenol'. It's just not the right place to be asking those questions. Initially, I was going to try and answer, but I don't even try and answer anymore because I think you just confused people.

The more knowledge the mothers acquired the better they were able to discern the best places to consult for the information they needed. Ivy ultimately summed up how the mothers experiences helped them determine the best places for information. She states, "As my questions became more clear the sources that gave me answers became more clear".

**We're the experts.** The mothers face many challenges with regard to providing the best health care to their child. This subtheme reveals the mothers must develop and possess a high level of health literacy due to the complexity involved in managing the disease. These complexities include interactions with a variety of medical and health care

specialists, interpreting medical diagnoses, providing medical/technical procedures (e.g., gastrointestinal feedings, tracheotomy care, etc.) to their child, understanding medical instructions/recommendations, and the lack of familiarity of many health care providers with Alagille syndrome. All of these elements must be addressed at a high level of knowledge and information/computer literacy, so the mother can engage in complex conversations with health care providers. Clare's statement represents almost all of the mother's experiences with developing their expertise in regard to how the Alagille impacted their child, "I remember reading the medical articles and then having to look up every other word because I didn't know what they were". In addition, Clare's statement reflects the determination and tenacity she felt was necessary to insure meaningful and helpful interactions with health care providers.

Regardless of the physicians' willingness and level to engage in learning about Alagille syndrome, the mothers still proceeded to gather information online to either assist/supplement or inform the doctor about Alagille syndrome. Elle's experience is typical of the mothers:

I got the information myself; her doctor didn't really, know or have any experience. So, I went online myself and found horrible information; so that didn't help anything at all, but I've notice the further we go along in this [journey with Alagille] the more I've educated myself on it, the less helpful they've [the health care providers] become.

As the mothers acquire more knowledge and hence, competence and confidence in managing their child's disease, they were likely to encounter doctors and other health care providers who were not interested in collaborating with the mother and treating her as a partner in the management of her child's disease. Faith states, "there have been a few times when I have been discounted; like my opinions and thoughts were discounted because I don't have a medical degree." Ann's excerpt illustrates the dismissal of the mother's knowledge and offers a plausible reason as to why this experience occurs:

I think that, I mean this was just my situation, but I think that applied to a lot of health care for parents of children with these rare diseases because they don't all fit into this one cookie cutter. You have to keep thinking outside the box and working things out and trying different things. Health care providers are busy and they've got so many other patients that the parents end up having to really advocate for their child and do the research on their own and bring it back to the health care provider. That's where it's important for the health care provider to be willing to hear what the parents are bring to them and [be] open to suggestions.

Faith further supports the need for health care providers to engage with the mothers of children with rare disease differently than mothers of children with more common chronic illnesses. In addition, Faith's statement reflects the fact maternal experience in managing her child's rare disease does not translate as valid and credible knowledge in a health care setting.



As a parent, because as a parent when you first get diagnosed, all you see are doctors and nurses. [That is] not to say in a bad way that they [doctors and nurses] don't understand, but they don't unless they have had a child with that. So they can say 'you need to do this and this and this' and uh, no that doesn't work actually, you're not, you're not listening to me.

#### **Theme Four: Seasons of Online Communication**

The mothers described in their narratives an experience that was best termed by Faith as "seasons". She noted that with Alagille syndrome "there are good seasons and there are bad seasons". Faith further explained seasons encompassed all the experiences families have with this disease where there are acute phases, medical procedures and lab tests and then there are periods in a child with Alagille's life where the disease is more stable. An excerpt from Faith's narrative illustrates this point:

It was just seriously like you were doing good. You'll have a good season or two and then boom here comes this. It's like "really one more thing" Okay; I can tend to get overwhelmed. That's why I am not going to listen to this voice [she tells herself]; it is not overwhelming, this is just something we are gonna look into [get information online and from health care providers] and we're gonna go with whatever it is. You start on a good roll for a season and then you feel like your feet get knocked out from under ya.

Faith's term captured best what all the mothers were trying to express in terms of the good and bad periods in their child's life. The mothers in this study shared that depending on the "season" their use and engagement in online communication may be more or less.

**Using online communication more.** The mothers described the need to use online communication more during a bad season. Examples of bad seasons were the initial diagnosis as well as pending labs, medical procedures or changes in routine. Ivy reports, "We get about 3 good weeks anytime we change her medicine and then it starts to get worse. So that's when I started doing more research with the Alagille on the internet and the, Alagille Syndrome Alliance the ALGSA". Clare described an interesting phenomenon experienced by mothers of children awaiting liver transplantation, one of the treatments for Alagille.

This is what needs to happen, um but, watching it's this weird thing when they're listed the sicker they are, the higher they are on the list, and has this very weird thing of almost wanting them to get sicker to move them higher on the list and that does funny things with your brain of, "wait do I want my child to be better or do I want them to be sicker?" So that, that was uh-- very challenging to try and navigate you know almost to the point of well you know well, if we don't give him the medication then he is gonna get sicker. Then you start playing these mental games with yourself. Online with the website, the organizational website, you type in there their lab numbers and you it gives their PELD score and so you play around. Well, if his INR goes to this then look at where his PELD score

goes and then you say wait a minute if his INR goes up to that then he would be really, really sick and so it's this really weird game that you play with yourself (chuckles) of wait minute you know.

Jan, another mother of a child who received a liver transplant reflected on her use of online communication during bad seasons. Jan notes, "I did notice that we were a lot hungrier for information when her symptoms were the very worst that was about age zero to four before her transplant; before she was feeling so much better".

**Using online communication less.** When times are better or a child is doing well or having a "good season" several mothers noted they use the internet specifically for information related to Alagille syndrome less. Mothers of older children noted that generally as the child got older these bad seasons were less frequent and their life with the disease stabilized. The older children were reported to enjoy less complicated and better health regardless of whether they received liver transplantation. Ann's narrative describes what many mothers experienced:

I do think that not all the time, but as the kids get older, their health kind of stabilizes and so for a lot of the kids it stabilizes and then your need for information and support becomes less and less. You're still connected, but I think those early years until about 8, 9, 10 are really tough years because there's still so much going on. I was just thinking [about] reaching out as much as you can to others and getting information and not trying to reinvent the wheel yourself

because it has already been invented. I see myself online it's less and less for the older he gets. It's not as all-consuming as it was when he was little.

Faith's mother also noted that she uses online communication less for Alagille. Faith states:

I am on that Facebook page, but I don't use it a lot. I have in the last probably, I'd say 5 years faded off of that [Alagille related activities]. Um, we just kinda transition to that to who Charity is and she is not as sick as she was. I don't have as many questions.

The mothers' report they did still enjoy relationships and support received from continued participation in the online community. However, these activities were consisted mostly of socializing and to provide advice and hope to parents new to the Alagille community, rather than seeking information.

### **Integration of van Manen's Life Worlds**

Van Manen (1990) suggested in using an interpretive hermeneutic approach, one seeks meaning in an experience. Identifying and describing the meaning of an experience involves "reflective understanding" (p. 86) and working on "mining meaning" (p.86) from the various reported lived experiences. One way to reflect and extract meaning from these experiences involves using a "wholistic reading approach" (p.93). Wholistic reading requires the researcher to examine the text in its entirety and then develop phrases or sentences that convey the main ideas of the larger body of text. In addition,

van Manen identified what he termed “lifeworld existentials” (p.101) to frame the reflection of the research process. The four fundamental lifeworld existentials described by van Manen were lived space (spatiality), lived body (corporeality), lived time (temporality), and lived other (relationality).

### **Lived Space**

Van Manen (1990) stated “lived space is felt space” (p.102) and is something most people do not reflect upon. Van Manen further described this concept of lived space referring “to the world or landscape in which human beings move and find themselves at home” (p.102). The actual home was central to the daily lives of these mothers and that of their child and the family. However, when applying this existential to this study the lived space must include consideration of cyberspace. Though cyberspace is not a physical space in the traditional sense, it is very much a “felt space” to the mothers of this study. Van Manen states “the space in which we find ourselves affects that way we feel” (p.102). Online communication and its increasing presence through mobile devices allows these mothers to experience the benefits and challenges of having an available and constant connection to others and, thus, affects how they feel.

Participants described the various lived spaces that impact their lives and triggered chronic sorrow and unpleasant feelings. The mothers’ narratives revealed online encounters with Facebook, You Tube, message boards and Google stirred up fears, sadness, anxiety and sensations of feeling overwhelmed. The accessibility of information and the ease of connection with others “is just easy”, but accompanying that lived space

was the realization that “you have hopes and expectations for their [the child’s] life. So, when you finally realize that your child has that mutation it is just hard to accept...the hard part, realizing that our life is gonna take a different turn”. It is in cyberspace that the participants found themselves most immersed when trying to understand and manage their child’s illness.

Cyberspace and the home were the main spaces that affected the mother’s feelings. However, in addition to cyberspace and home, these mothers often had to contend with institutional spaces such as hospitals, numerous health care provider offices, laboratories and radiology departments. Hope summed up the experience of many of the participants when she discussed feelings that were triggered by an institutional space, specifically nuclear medicine: “when we[child had the MRI] had to have a cardiac MRI at one point... it was a sedated cardiac MRI I remember waiting there for her which is difficult to begin with” (voice breaks). The variety and complexity of the Alagille syndrome placed the mothers in many places that had the ability to trigger chronic sorrow and unpleasant feelings.

### **Lived Body**

The existential of lived body as described by Van Manen (1990) proposes that “[i]n our physical or bodily presence we both reveal something about ourselves and we always conceal something at the same time [...]” (p.103). The narratives of the participants viewed within this lifeworld manifested through verbalizations of their emotions, feelings and physical connection and awareness of their child’s health. These

mothers reported the rarity of the condition often caused health care providers unfamiliar with Alagille syndrome to overlook or dismiss what could be critical symptoms of findings in their child. These participants were highly attuned to the most subtle changes in their child's health. The mothers reported the highly variable nature and expression of the disease led them to be vigilant in using online communication to manage their feelings and increase their knowledge to advocate for their child so best outcomes were achieved.

### **Lived Time**

Van Manen(1990) states that lived time is “subjective time” or “temporal time” (p. 104) that encompasses the past, present and future. Van Manen further describes this lifeworld by noting the past, “now sticks to me as memories or as (near) forgotten experiences that somehow leave traces on my being – the way I carry myself (hopeful or confident, defeated or worn out), [...]” (p. 104). The participants in this study provided evidence of the influence of lived time in their narratives. The mothers noted in the early days and weeks of after their child's diagnosis the experiences, feeling, events and interactions with health care providers left a profound mark upon them. Reeling from the shock of the diagnosis, mothers of older children pointed out that their online communications did not provide immediacy they sought in managing their feelings, obtaining information and developing support. As online communications developed, these mothers along with mothers new to the community experienced the ability to find much more information, developed online relationships through social media and built

practical parenting tips of children with Alagille syndrome into their repertoire of knowledge.

Van Manen continues to further elucidate lived time by noting that “[t]hrough hopes and expectations we have a perspective on life to come [...]” (p.104). This quote is particularly notable and helpful in describing the meaning and importance of online communication in helping these mothers to manage their unpleasant feelings. The mothers’ narratives consistently touched upon the fact “each Alagille kiddo is different” and the presentation of the disease varied widely. These mothers were able to find positive affirmation and hope for their child’s future through information and relationships that developed as a result of being engaged with online communication. However, this same form of communication reported by the mothers could sometimes trigger angst, fear and sorrow with regard to the uncertainty and unpredictability of the disease resulting from things they read online about other children.

### **Lived Other**

The existential of lived other is described by van Manan (1990) as “the relations we maintain with others in the interpersonal space we share with them” (p.104). In this context, van Manen proposes humans search for the meaning of “other” (p.104) in the sense of life’s purpose and the meaningfulness in a lived experience.

The narratives of the participants revealed the mother’s protective nature and the need to seek resources that could aid them in this task. This includes the support derived from online communications as well as the development of support in online



communities. In addition, these mothers revealed the ability of increase knowledge and connect with other moms of children with Alagille syndrome as a result from engaging in online communication was necessary to support familial relationships (i.e. spouse, other children). Several mothers, who mentioned during the interview they had other children, discussed having a heightened awareness and sensitivity to include their children in family decisions regarding issues involving the child with Alagille syndrome and the need to create special time for them. In general, these mothers experience enhancement in family relations by the development of their proficiency and knowledge in the care of their child with Alagille syndrome. This increase in ability and confidence and ability to protect and advocate is in large part due to the information and relationships they have attained from use of online communication.

### **Summary**

This chapter provided a detailed description and analysis of the experiences of sixteen mothers of children with Alagille syndrome in using online communication to manage unpleasant feelings including chronic sorrow. One overarching theme and four essential themes were revealed during the analysis of the mothers' experiential narratives. The overarching theme identified "Online Communication is Essential to a Rare Disease Community" was predominant in the narratives of the mothers as they shared their lived experiences of attempting to find information and support to assist them with their journey in caring for a child with the rare condition of Alagille syndrome. The four essential themes: 1) connectedness, 2) online triggers, 3) empowerment, 4) seasons of online use, provided a lens by which the researcher could describe the mothers'

experiences resulting from the wholistic readings conducted by the researcher. Lastly, the researcher integrated the study findings within van Manen's "lifeworld existentials". In Chapter 5, the essential themes are discussed as they relate to existing research and the implications for nursing practice, education, research and policy.

## **CHAPTER 5**

### **Themes, Implications and Conclusion**

This chapter discusses the overarching and essential themes discovered during the analysis of the textual data and relates the themes to previous research and to the conceptual framework of chronic sorrow described in Chapter 2. In addition, this chapter discusses recommendations for practice, education, research and policy with the goal of improving experiences for mothers of children with rare diseases such as Alagille syndrome.

#### **Return to Research Questions and Assumptions**

The researcher sought to explore the lived experience of mothers of children with Alagille syndrome in using online communications to manage chronic sorrow. The main question for consideration was developed using the theory of chronic sorrow as a guide in exploration of the experience of chronic sorrow in mothers of children with Alagille syndrome. Additional questions for consideration included: What forms of online health communication are used by mothers of children with Alagille syndrome and what are their experiences with online health communication? What chronic sorrow experiences are reported by mothers of children with Alagille syndrome? The discussion of the data

and development of the overarching and essential themes focuses on these questions and assumptions.

### **Themes**

Research studies build the knowledge base of a discipline. However, van Manen (1990) cautioned researchers that studies that “do little more than present and organize transcripts fall short of their interpretive and narrative task” (p. 167). Munhall (2012) extended this line of thinking when she suggested the significance of phenomenology is “stating the implications for change that emerges from the interpretations we glean from our participants on the meaning of various experiences” (p. 121). She further notes results of studies using phenomenology can be used for guiding development of recommendations of changes in practice and policy. According to Munhall (2012), phenomenological studies can result in “increasing our capacity of care and compassion, emancipation from oppression, and raising our consciousness of what was not known or otherwise erroneous” (p.121).

This study attempts to describe how online communication is used by mothers of children with the rare disease Alagille syndrome to manage chronic sorrow. As noted previously, research on how mothers use online communication for rare conditions is limited, while research about coping behaviors in mothers of children with chronic illnesses is extensive. While researchers are beginning to focus somewhat more on the uniqueness of coping with rare chronic diseases, research studies could not be located that specifically looked at online communication in the management of chronic sorrow.

This study attempted to view online communication by mothers use within the context of chronic sorrow framework. The researcher found that about half of the mothers did not experience chronic sorrow. These mothers revealed feelings which can be better termed unpleasant feelings. Regardless of whether the mothers identified unpleasant feelings or chronic sorrow, their lived experiences with online communication reflected commonalities, which allowed discovery of an overarching theme and four essential themes.

The mothers' narratives revealed the overarching theme that online communication is essential to a rare disease community in the management of chronic sorrow or unpleasant feelings. In addition, four essential themes, connectedness, online triggers, empowerment and seasons of online communication, contributed to the overarching theme and provided a more comprehensive description of the lived experience of the mothers.

Online communication has profoundly impacted the rare disease community. For mothers in this study, the diagnosis of their child with a rare disease became a stressor that led them to engage in online communications early in their journey with Alagille syndrome. Their use of online communications was described as helping manage their chronic sorrow/unpleasant feelings or heightening these emotional experiences. The rarity of the condition led the mothers to continue to engage in online communication "even if the information being sought could prove scary and increase their anxiety" (Gundersen, 2010, p. 92). The mothers felt alone, isolated, and unable to rely on their

health care providers for information and guidance, as few providers had even heard of the disease. Therefore, online communication was necessary for the participants to gain the knowledge and expertise in managing the disease and educating others including health care providers which was similar to findings revealed by Leonard and colleagues (2004). In addition, consistent with research by Nordfeldt and colleagues (2013), the perceived seriousness of the situation affected the mothers' choice and desire to use online communication.. The mothers' narratives voiced a strong need to meet others with the same disease and similar experiences which was consistent and well documented in studies of parents with chronically ill children (Baum, 2004; Coulson & Greenwood, 2011; Han & Belcher, 2001; Hodges & Dibbs, 2010; Leonard, et al., 2004; Margalit & Raskind, 2009; Tozzi et al., 2013). The participants longed for understanding and knowledge. Over time, the mothers' development of their expertise on Alagille syndrome allowed them to manage emotions, develop knowledge and, thereby, a sense of control of the situation that ultimately put them in the position to be their child's advocate (Gundersen, 2010). The mothers all enjoyed being a part of the online community. However, many participants opined that while belonging to an online community filled the same need of meeting others in similar circumstances; it could not completely replace the feeling of face to face interactions.

### **Connectedness**

Connectedness encompasses the importance of emotional support found in online communication, particularly in support groups and social media sites like Facebook. It

was important for mothers to find and connect with others mothers and parents of children with the same illness. Consistent with the literature, emotional and informational support were used by the participants (Coulson & Greenwood, 2011). Several studies have provided evidence of the value of the emotional component of online parent-to-parent support (Baum, 2004; Coulson & Greenwood, 2011; Gundersen, 2010; Han & Belcher, 2001; Leonard et al, 2004; Nordfeldt et al., 2013). All mothers felt online support was critical to helping them manage chronic sorrow or unpleasant feelings and increase their knowledge of Alagille syndrome. Obtaining information about their child's disease was essential for understanding and adjusting to an initially distressing situation (Gundersen, 2010). In time, several mothers of older children evolved from initially being supported to being able to support others. This cyclical process was described in a qualitative study of parents by Kerr and McIntosh (2000), in which the authors suggested this activity likely leads to empowerment.

The participants in this study evolved with the changes in technology and the internet. Online communication changed the type and frequency of the mothers' communications. Online communication evolved, allowing the mothers to seek support, information and form a sense of connectedness from all over the world. Many people who are in stressful situations participate in web sites offering online support (Margalit & Raskind, 2009). Most of the mothers in this study began their experience of feeling connected by using social media, specifically Facebook. Social media replaced asynchronous formats for online communications with other parents, often providing

immediate responses from users. This finding highlights a complaint of the limitation of message boards and list serves in which users expressed disappointment at the lack of responses to posted messages (Coulson & Greenwood, 2011).

The majority of the mothers used Google as their search engine for the initial inquiry for information and resources, but the mothers also used other websites such as Yahoo and Web MD. This finding was consistent with other studies in which individuals seeking health information often first visit general search engines because they were unfamiliar with or unaware of online data bases (Roche & Skinner, 2009; Semere et al., 2003; Yi, Stvilia, & Mon, 2011). Over time the mothers were able to improve their ability to find information and assess its relevance and trustworthiness. Gundersen (2010) suggested online communication “enables people with different information gathering preferences and skill levels to increase their understanding of a difficult life situation by allowing them to manage their different informational needs” (p.92).

### **Online Triggers**

While the benefits of online communication far outweighed perceived risks, these mothers acknowledged it sometimes acted as an online trigger for unpleasant feelings and chronic sorrow. In the theory of chronic sorrow, identified trigger events in the literature include developmental milestones, special events, and certain activities or situations that reflect the disparity between the parents’ “ideal” and the reality of their chronically ill child (Eakes, Burke, & Hainsworth, 1998). These triggers consequently result in a resurgence of chronic sorrow.



The mothers identified both traditional triggers and online triggers. The traditional triggers, such as their child being diagnosed with a new condition, the development of a new symptom or a pending procedure, are consistent with findings by Roche and Skinner (2009) in parents of children referred for genetic services. However, this study also identified specific online triggers that caused the mothers to report feelings of unpleasantness that included chronic sorrow. These online triggers included learning about the death of a child with Alagille syndrome and exposure to excessive medical information and social media experiences.

Online information can be ineffectual for managing unpleasant feelings and chronic sorrow if it results in information overload. Several mothers in this study expressed the overwhelming nature of various online experiences. The ease of accessing health information may make parents feel compelled to use the internet (Ziebland, 2004). However, consistent with the research, exposure to too much information caused mothers to experience an increase in anxiety (Tozzi et al., 2013). Parent exposure to excessive online communication appears to hamper the ability to judge the amount of information the parent actually needs because the parents' world has been so significantly disrupted (Gundersen, 2010).

The mothers in this study risked information overload because most health care providers were unfamiliar with Alagille syndrome and, thus, provided inadequate information. The providers' lack of knowledge led the mothers to engage in significant online communication almost immediately after their child was diagnosed. This finding

is consistent with research by Leonard and colleagues (2004) on parents of children with rare neurologic disorders and by Tozzi and colleagues (2013) on Italian parents of children with rare disease. The rarity of the illness is a critical and defining factor with regard to parents engaging in heavy online communication. Gage and Panagakis (2011) did not find that parents of children with pediatric cancer used the internet in the acute phases of diagnosis to obtain health information, preferring to rely on their health care provider and deeming the internet as “untrustworthy and frightening” (p.455). This critical difference seems to highlight important dimensions to consider in online communication of mothers of children with rare and chronic conditions as well as serious and potentially life-threatening health conditions.

### **Empowerment**

Empowerment by the mothers developed as they amassed information and acquired knowledge, predominantly through online communication resources. This finding was important in that Dellve and colleagues (2006) noted in their prospective study of stress and wellbeing in parents’ of children with rare disease that developing parents’ competence and empowerment might prevent stress related to parental incompetence. In addition, empowerment development in the context of the use of online communication for health information was revealed as prominent in several studies (Broom, 2005; Dolce, 2011; Ziebland, 2004)

Over time, the mothers learned the best sites to go to for the information they needed. The mothers were exposed through networking with other parents to specific

sites for useful and trusted information regarding health and medical information on Alagille syndrome. Specifically, the practical advice offered by networking in the online community contributed to empowerment by provision of information and support, which is consistent with findings described in mothers of children with learning disabilities and attention deficit hyperactivity disorder who used online communication (Margalit & Raskind, 2009).

The mothers developed their expertise in managing Alagille syndrome as a direct response to not being able to rely on the health care provider as the predominate provider of information in the disease. Dolce's (2011) research on cancer survivors and caregivers described similar motivations. Dolce described "disenchantment" with health care relationships related to informational support as one of the factors that drove patients and caregivers to conduct internet searches for health information and resources.

Once the mothers were knowledgeable, many expressed their expertise was discounted and/or not valued by the health care professionals. This finding was similar to Swallow and Jacoby's (2001) research of mothers' of chronically ill children where mothers' voices were "unheard" until they developed their own strategies for communicating and negotiating with health care professionals. In addition, an important characteristic of empowerment cited in the literature was parental participation in the health care of children with chronic illness (Resendez, Quisti, & Matashzi, 2000). Resendez and colleagues (2000) suggested it was necessary to support and consult parents as experts in the care of their child as well as develop parents' knowledge, skills,

and self-efficacy so the parents can provide care to their child and manage situations that could invoke stress and compromise their emotional health.

Empowerment also occurred when the mothers controlled the amount of information communicated in updates about their child's disease via social media, thereby protecting their emotions and potentially their child's esteem. The control of online information reflected a sense of power in the online community which was similar to findings by Itzhaky and Schwartz (2000) in which they described a sense of power in the community realm developed in response to the individual's feeling of control over external forces.

### **Seasons of Online Communication**

The mothers' online communication and information seeking behaviors changed over time based upon their child's development and illness trajectory. The participants predominantly used online communications for informational and emotional support. Information and emotional support were most often the two main types of social support cited in connection with online social support research (Coulson & Greenwood, 2011; Han & Belcher, 2001; Leonard et al., 2004; Margalit & Raskind, 2009). Mothers of children of older children with Alagille syndrome used online communication less for information on Alagille syndrome and more for providing support and information to others new in the journey with Alagille syndrome. However, if the child experienced a new symptom or was scheduled for a procedure or test, the mothers' engagement in information seeking online increased. Mothers of younger children with Alagille

syndrome also reported less use of the online communication when they child was more stable or had less symptoms.

The mothers' choice and level of frequency in the use of online communication were based on previous experience and knowledge about what matched their situation. This theme is consistent with research by Nordfelt and colleagues (2013) conducted in parents of adolescents with type 1 diabetes mellitus in which the researchers suggested that parents' information seeking related to their life situation, their child's development, and the different phases of illness. Gundersen (2010) also noted in her study of Norwegian parents of children with a rare disease that most of the parents over time reduced their information seeking activities.

One can view these behaviors through the lens of the theory of optimal matching (Cutrona & Russell, 1990), which focuses on the individual's perceived controllability of a situation. Cutrona and Russell (1990) proposed that individuals are likely to view emotional support as being more helpful when they are experiencing distressing circumstances that they perceive are out of their control. In contrast, individuals may consider informational support to be more useful when they feel that they can use it to control the situation. Optimal matching theory may explain the participants' reports of variation in online communication use based upon the disease trajectory. The mothers may feel some aspects are beyond their control (e.g. new diagnosis, procedure/test) while other aspects remain in their control (e.g. managing itching, parenting, responding to comments from others on their child's appearance).

## **Implications**

These findings serve to assist in the development of future research areas and help to better describe the phenomenon of online communication in management of unpleasant feelings among mothers of children with rare diseases. This section provides suggestions for improving online communication and general support for mothers of children with Alagille syndrome via practice, education, research and policy implications.

### **Implications for Practice**

The mothers' experiences in this study suggested that these mothers did not perceive themselves as getting the typical support and information afforded to mothers whose children were diagnosed with more common chronic conditions. The complexity of the disease coupled with the unfamiliarity of the diagnosis among the many health care providers involved in the care of their child led these mothers to explore online communication almost immediately. Findings suggest health care providers need to advance their understanding that a diagnosis of a rare disease requires special care in guiding mothers with managing the unpleasant feelings and chronic sorrow associated with this news. In particular, providers need to assist mothers with accessing appropriate and helpful online communication.

Physicians, clinical nurses, advance practice nurses, and all other members of the health care team have the ability to greatly influence the early experiences of mothers who find themselves in this situation. The mothers of children with Alagille syndrome

expressed the need for guidance in understanding what this diagnosis means for their child and family, as well as where they can get the best and most helpful information. The paucity of information on rare diseases like Alagille syndrome requires clinical health care providers be familiar with and have knowledge of the most reliable sources for information, which includes relevant and helpful online communication sources (e.g. websites, online communities, data bases, etc.). Additionally, health care providers have a duty to develop in their patients' parents a sense of awareness of strengths and limitations to engaging in the online community.

The health care landscape has changed and individuals come to office visits armed with information they obtained online. Increasingly, individuals view themselves as active partners in their health care decision making (Boyer and Lutfey, 2010; Ayers & Kronenfeld, 2007; Leonard et al, 2004). This dynamic affects the relationship health care providers have with their patients (Zieband, 2004), thus necessitating a change in approach. During office visits and hospitalization, clinical health care providers should regularly ask about online communication the mother has used so inaccuracies can be corrected and accurate information can be reinforced. Health care providers should consider assessing mothers for information/computer literacy. Health care providers should consider a more active role in online communities as this would help to support accurate and valid information. An interest in the types of health information the mothers seek, as well as the places online where they seek it, can help validate the mothers' role in obtaining information online. An inquiry made to the mother creates a situation where

the clinician can guide the mother to better resources by suggesting relevant key words, data bases, and websites. In addition, discussing with the mother the websites they use can help health care providers understand the types of information mothers are interesting in learning and, thus, help the provider anticipate the informational needs of mothers.

The interviews in this study allowed the researcher to receive direct recommendations from the mothers as to how health care providers could support the emotional needs of parents of children with Alagille syndrome. Their complied suggestions are listed below.

- Provision of a health care professional similar to a social worker to assist with the emotions experienced at the time of the diagnosis (the parent should not have to request this service, but it would be automatically included)
- “Recognize when we are struggling and help us” (via provision of compassion, empathy, information, consideration, knowledge)
- Taking an interest in the disease and helping the parents “figure things out and learn about it”
- Help the parents find an online community
- “Respecting how much mothers know”

### **Implications for Education**

The Institute of Medicine (2011) report on the future of nursing emphasizes the need for nurses to shift in thinking and practices. One of the emerging 21<sup>st</sup> century health care challenges is the number of greater number of people with chronic conditions,



resulting from increasing capabilities of health care systems to treat chronic illnesses. In addition, nurses need to re-conceptualize their roles as “care coordinators, health coaches and system innovators” (p.67) to meet higher quality care standards and delivery of more care in community settings.

The training and philosophy of nurses can be beneficial in addressing online communication use with regard to the chronic sorrow and unpleasant feelings in mothers of children with rare diseases. Nurses are taught to treat patients from a disease management perspective, however nurses are also taught to consider the patient from a psychosocial, spiritual and family/community perspectives. In addition, providing care for underserved populations in community settings has long been a major goal of nursing. Mothers of children with rare diseases, while not underserved populations in the traditional sense, are very much underserved and are deserving of the attention afforded to the traditionally underserved. The mothers in this study shared the emotional rollercoaster of chronic sorrow and unpleasant feelings and how their use of online communication had the potential to calm or heighten these feelings. These mothers did not report any guidance from nurses or other health care providers in navigating online communication formats to assist them with addressing their concerns. Curriculum content in all nursing programs should emphasize better awareness of existing information resources, the need to assist families in identifying reputable websites/support resources online, and enhancing awareness of telemedicine, e-health formats and other new technological resources to support care. This suggestion is

consistent with *The Essentials of Baccalaureate Education for Professional Nursing Practice* (American Association of Colleges of Nursing, 2008) which notes curricula should include virtual learning experiences like immersion in online communities or moderating online support groups.

Nurses in community health care settings need to become more proficient with technology and embrace it and the value it brings in expanding health care in the community. The ability to guide mothers of children with rare diseases to the most appropriate online communication resources is just a start. As cited in Chapter 1 and in the narrative of the mothers of this study, parents dealing with rare diseases often do not have access to health care providers who are knowledgeable about their child's condition and, thus, find it difficult to obtain health information regarding the specific condition afflicting their child. Oftentimes, parents must travel great distances to large academic medical centers to get the most knowledgeable physicians/specialist and most current treatments for their child's condition. Research emerging in this area suggests that telehealth services provided by nurses and brought to parents of children with special health care needs may increase access to care for those in rural and medically underserved areas at no additional costs (Hooshmand, 2010). The research also indicates that parents' perceptions of the system of care were reported as more positive (Hooshmand, 2010). Nurses and all health care professionals must continue to explore and implement technology strategies to compliment care that promotes optimal outcomes for children and their families.

## **Implications for Research**

Researchers have begun to discern subtle differences in how online communication use can vary among mothers of children with more common chronic illnesses and mothers of children with rarer chronic diseases. The emotional/psychosocial response within the context of use of online communication also tends to vary between these two groups. The findings in this study suggest additional research should be conducted regarding the follow up of mothers after the diagnosis of their child to determine whether they have connected with resources online.

Current research in the area online communication and coping in the context of chronic illness is substantial, but it remains limited with regard to mothers of children with rare disease and in related coping areas like chronic sorrow. More research also is needed on how fathers of children with rare disease and adolescents and adults with rare disease use online communication, the impact of online support, and the adaptation, coping and feelings they have experienced over time. In addition, the development of assessment tools that allow for quick identification for the normal emotions of chronic sorrow and unpleasant feelings would help clinicians provide appropriate resources such as online communication/support.

The ability to obtain information and increase knowledge is essential for patient advocacy and the parent's ability to advocate for their child. Additional research is needed to explore when an individual may seek information online for some health related experiences as one of the theme of seasons led the researcher to identify internet

use varies over time and circumstance. In addition, mothers and families armed with information obtained from online communication often feel more empowered.

Consequently, research that examines the impact of online communication on the dynamics of the health provider relationship or consultation in the context of rare disease would also be beneficial.

### **Implications for Policy**

With regard to policy implications, the rare disease community must be viewed within the context of an organizational level. Individually, rare disease organizations are small and meaningful only to those individuals affected by the particular condition. However, collectively rare disease organizations wield great power and have the ability to influence the political agenda. The National Organization for Rare Disorders (NORD), a U.S. based resource for anyone affected by rare disease, has as its motto “Alone we are rare. Together we are strong.” NORD, along with the individual rare disease organizations, seeks to “provide patients and their families with programs of education, advocacy, research, and service” (NORD, 2013).

Rare disease organizations are used to fighting for recognition and improved care for those affected. Improved care encompasses research on psychosocial issues that may impact individuals with rare diseases and their families. Psychosocial issues and emotional health are becoming increasingly important in the care of those with chronic illness. When the chronic illness is rare, this further complicates those experiences and feelings. The mothers in this study described the chronic sorrow and unpleasant feelings

they faced and how online communication, most often a first line resource, was vital to managing their feelings, regardless of the disadvantages or risks they encountered.

Unlike mothers of children with more common chronic diseases, the health care provider/clinician was often not a first line resource. Health care organizations need to collaborate with rare disease organizations and/or NORD to develop guidelines for the use of a wider range of communicative technology such as videoconference (e.g. Skype), structured chat, and health oriented social media platforms. These options may facilitate communication among health care providers and mothers, which would ultimately support and benefit mothers' emotional/psychosocial health and enhance their knowledge.

### **Conclusion**

The personal experiences of the researcher led her to study how online communication was used by mothers of children with Alagille syndrome in managing chronic sorrow. Her experiences and the research literature guided her interest in exploring mothers' descriptions of experiences of chronic sorrow. This study offered mothers the opportunity to speak about their experiences of using online communication in managing psychosocial issues. It is hoped by describing the experiences of the mothers in using online communication to manage chronic sorrow and other unpleasant feelings that health care providers will better recognize and understand how they can provide support and resources to mothers of children with rare diseases like Alagille syndrome.

A hermeneutic phenomenological approach, guided by van Manen (1990) was used to reveal the lived experiences and interpret the meanings of the experiences. While chronic sorrow as defined by Eakes, Burke and Hainsworth (1998) was not evidenced in a majority of the participants, all mothers' use and experiences with online communication were very similar. The rarity of the disease influenced these mothers' experiences and engagement in online communication. The mothers' use of online communication was found to differ from mothers of children with more common chronic diseases. The analysis of the mother's narratives revealed one overarching theme and four essential themes, which was discussed in the context of the current research literature. This chapter also discussed the implications for practice, education, research, and policy.

## APPENDIX A

George Mason University

APPROVED  
8/4/16  
George Mason University

Mother's Use of Support Networks

### **Mothers of Children with Alagille Syndrome (ALGS) Wanted for a Research Study**

This research study is being conducted by a nurse practitioner and PhD nursing student as part of her doctoral dissertation. The purpose of this study is to describe how mothers use support networks. In addition, the researcher wants to describe the mother's thoughts, feelings, and experiences in caring for a child with a rare disease.

Subjects should be mothers who are actively responsible for the care of their child with Alagille Syndrome. Participants need to be 18 years of age and older and be able to speak, read and write in English.

There are no benefits to you as a participant, other than to further research in the area of support and advocacy of mothers of children with rare diseases such as Alagille syndrome. Findings may help nurses and other health care providers provide more effective support and also design effective interventions that may help mothers of children with rare diseases.

To learn more about this research, contact [Adriana Glenn RN, MN, MA, FNP-BC, PhD \(c\)](#) by email @ [aglenn2@gmu.edu](mailto:aglenn2@gmu.edu) or call 703.426.2062 or 703.994.7346 (cell).

This research is conducted under the direction of Dr. Charlene Y. Douglas, Associate Professor, School of Nursing/College of Health and Human Services.



5/6/2013

## APPENDIX B

May 6, 2013

Dear Online Community Member,

I am a PhD nursing student conducting my dissertation study that will describe the support systems, thoughts, feelings and experiences of mothers of children with Alagille Syndrome (ALGS). Currently, I am trying to recruit mothers who might be interested in participating in this important research study. Alagille syndrome and its impact on mothers is very important to me and dear to my heart because my daughter Avery had this disease. Findings from this study may help guide nurses and other health care providers in providing information on support and advocacy for parents struggling to cope with rare diseases, including ALGS. Please let me know if you can help. I look forward to hearing from you.

Sincerely,

Adriana Glenn RN, MN, MA, FNP-BC, PhD (c)

George Mason University

APPROVED  
8746  
George Mason University



## APPENDIX C

### *Mothers of Children Diagnosed with Alagille Syndrome: A Description of Support and Thoughts, Feelings and Experiences*

#### **INFORMED CONSENT FORM**

##### **RESEARCH PROCEDURES**

This research is being conducted to describe support systems and thoughts, feelings, and experiences of mothers of children with Alagille Syndrome (ALGS) that have helped the parents navigate this complex disease and advocate for their children. If you agree to participate, you will be asked to take part in an interview(s) conducted by telephone or Skype (your choice).

There will be two parts to this interview. The first part is designed to gather some basic background information about you and your child. Part one will not be audio-recorded, but the researcher will document the responses on a form. The second part involves questions related to experiences with health communication, seeking information and some of your thoughts/feelings since your child was diagnosed with Alagille syndrome. The second part of the interview will be audio-recorded with your permission. The duration of the interview would be approximately 60 minutes.

##### **RISKS**

There are no foreseeable risks for participating in this research.

##### **BENEFITS**

There are no direct benefits to you as a participant. Your participation may help research in the area of support networks and advocacy of ALGS parents and their families. Findings may help guide nurses and other health care providers in providing information on support and advocacy for parents struggling to cope with rare diseases, including ALGS.

##### **CONFIDENTIALITY**

Participants' full names, locations, dates of birth or other participant identifiers will not be used in the study in any form, including notes, articles, etc., other than to create a key linking real names to a pseudonym that will be used at all times. The key that links the actual names to the pseudonym will be stored in a locked cabinet separate from the rest of

the data in the study to which only the researcher has access. Upon completion of the study, transcribed data/notes/audiofiles and the key will be destroyed three years after completion of the study.

**PARTICIPATION**

Your participation is voluntary, and you may withdraw from the study at any time and for any reason. In addition, during the interview, you do not have to answer any interview question that you do not want to answer. If you decide not to participate or if you withdraw from the study, there is no penalty or loss of benefits to which you are otherwise entitled. There will be no costs to you or any other party.

**CONTACT**

This research is being conducted by Adriana Glenn, a student in the Department of Nursing at George Mason University. She may be reached at 703.426.2062 or 703.994.7346 (cell) for questions or to report a research-related problem. Her faculty advisor is Dr. Charlene Y. Douglas at 703.993.1937. You may contact the George Mason University Office of Research Integrity & Assurance at 703.993.4121 if you have questions or comments regarding your rights as a participant in the research.

This research has been reviewed according to George Mason University procedures governing your participation in this research.

**CONSENT**

I have read this form and agree to participate in this study.

\_\_\_\_\_ I agree to audio recording of the interview.

\_\_\_\_\_ I do not agree to audio recording of the interview.

\_\_\_\_\_  
Name

\_\_\_\_\_  
Date of Signature

Version date: May 16, 2013

## APPENDIX D

### Qualitative Interview Guide

Assigned Pseudonym:

Child's First Name:

Interview Date:

Mode of Interview: Phone/Skype

Start Time:

End Time:

Researcher introduces self and study

Consent form - review procedures, confidentiality, voluntary, can skip/stop at any point

Appreciation for participation

Introduction to the Interview

*There are two parts to this interview. The first part is designed to gather some basic background information about you and your child. Part one will not be audio-recorded, but the researcher will document the responses on a form. The second part involves questions related to experiences with health communication, seeking information and some of your thoughts/feelings since your child was diagnosed with Alagille syndrome. The second part of the interview will be audio-recorded.*

*The information on this page is NOT audio-recorded*

## Part 1: Background Information

### MOTHER INFORMATION

Information Type	Response
Mother's Age	
Mother's Race/Ethnicity	
Marital Status	
Highest Level of Education	
Employment	
Country of Residence/State	

### CHILD INFORMATION

Information Type	Response
Child w/ALGS Age	
Age when child dx	
Other dx	

*Audio-Recording Starts at this Point*

## Part 2: Health Communication, Information Seeking, Thoughts and Feelings Experienced Since the Alagille Diagnosis

I would like to ask you some questions related to how you get your information and some of the thoughts and feelings you have experienced since (name) was diagnosed with Alagille syndrome. I am interested in learning about your experiences and point of view so nurses can become more sensitive and helpful to mothers who have children with rare diseases.

1. (Assigned Pseudonym), when you first learned your child had Alagille syndrome, how did you feel about this news? (Probe: *What went through your mind?*)
2. How have you gotten help to deal with these feelings you described? (Probes: *What did you try/how did it work? Any people in particular helpful or not helpful? Who were they? Can you recall what they did that helped you or what they did that was not helpful? Did you use online sources to locate these people? Could you describe the process/experience of connecting with online sources?*)

3. How have you gotten your information on Alagille syndrome? (Probes: *What sources do you use? Can you give me examples? Do you use any social networking sites? Do you read blogs? Do you blog? Do you email?*)
4. Describe your experiences with these forms of online communication (follow up question to mother report of internet use) (Probes: *would encompass how these communication forms are used and if the mother reports, good and/or bad experiences*).
5. Thinking back to how you reacted at first to the news of (name's) condition, can you describe a time since when something happened and you had those same feelings of (*use individual's words in her response to #1*) all over again?
6. What feelings do you have right now when you think about (name's) condition?
7. Some caregivers say certain events tend to bring up these feelings (*whatever term they used in response to item #1*) again. Describe the events that may stir up or generate these feelings. (Probes *may include asking the mother to describe any developmental delays, special events or circumstances that triggered these feelings; In using online resources, could you describe any circumstances where an encounter online brought up these feelings?*) Were there other times when you had these feelings? (*If yes: Can you tell me about at least one of these times?*)
8. Were there other people aware that you were having these feelings? How did they know?)(Probes: *Were any of these people from your online community? Describe your experience (frequency amount of time, format) communicating your feelings with people from the online community*)
9. I hope that my study will help us give really practical advice to people who are providing care for individuals with Alagille syndrome. Is there anything that you would tell people about helping mothers like yourself? What would you tell them they can expect? What will they need to know? (Probes: *If you were asked to give advice on using online resources in caring for a child with Alagille syndrome, what would you share about using online communication?*)
10. Is there anything we have not discussed that you think is very important to tell nurses or other health care professionals about helping mothers like yourself?

## REFERENCES

- A Dictionary of Nursing (2008). Retrieved from <http://www.encyclopedia.com/1062-developmental-milestones.html>
- Ahlstrom, G. (2007). Experiences of loss and chronic sorrow in persons with severe chronic illness. *Journal of Clinical Nursing, 16*(3A), 76-83.
- Ainbender, J.G., Blanchard, L.W., Singer, G.H., Sullivan, M. E., Powers, L. K., Marquis, J. G., & Santelli, B.. (1998). A qualitative study of parent-parent support groups for parents of children with special needs. *Journal of Pediatric Psychology, 23*(2), 99-109.
- Allen, D., & Marshall, E. S. (2010). Spirituality as a coping resource for African American parents of chronically ill children. *Maternal Child Nursing, 35*(4), 233-237.
- Allen, M. N. & Jenson, L. (1990). Hermeneutical inquiry, meaning and scope. *Western Journal of Nursing Research, 12*(2), 241-253.
- American Association of Colleges of Nursing (2008). The essentials of baccalaureate education for professional nursing practice. Retrieved from: <http://www.aacn.nche.edu/education-resources/baccessentials08.pdf>
- American Psychological Association (2012). Understanding chronic stress. Retrieved from <http://www.apa.org/helpcenter/understanding-chronic-stress.aspx>
- Ayme, S., Kole, A., Groft, S. (2008). Empowerment of patients: Lessons from the rare diseases community. *The Lancet, 371*, 2048-2051.
- Ayers, S. L. & Kronenfeld, J. J. (2007). Chronic illness and health seeking information on the internet. *Health, 11*(3), 327-347.
- Baum, L. (2004). Internet parent support groups for primary care givers of a child with special health care needs. *Pediatric Nursing, 30*(5), 381-401.

- Bettle, A. M., & Latimer, M. A. (2009). Maternal coping and adaptation: A case study examination of chronic sorrow in caring for an adolescent with a progressive neurodegenerative disease. *Canadian Journal of Neuroscience Nursing, 31*(4), 15-21.
- Birch, D. A. (1998). Identifying sources of social support. *Journal of School Health, 68*, 159-164.
- Boyer, C. A. & Lutfey, K. E. (2010). Examining the critical health policy issues within and beyond the clinical encounter: Patient-provider relationships and health seeking behaviors. *Journal of Health and Social Behavior, 51*(1), S120-132.
- Bouchard, K. (1998). Issues in case management: Respite care. *Pediatric Nursing, 24*(3), 260.
- Bowes, S., Lowes, L., Warner, J., & Gregory, J. W. (2009). Chronic sorrow in parents of children with type 1 diabetes. *Journal of Advanced Nursing, 65*(5), 992-1000. doi: 10.1111/j.1365-2648.2009.04963.x
- Bragadottir, H. (2008). Computer-mediated support group intervention for parents. *Journal of Nursing Scholarship, 40*(1), 32-38.
- Brazil, K., & Krueger, P. (2002). Patterns of family adaptation to childhood asthma. *Journal of Pediatric Nursing, 17*(3), 167-173.
- Broger, B., & Zeni, M. (2011). Fathers' coping mechanisms related to parenting a chronically ill child: Implications for advanced practice nurses. *Journal of Pediatric Health Care, 25*(2), 96-104. doi: 10.1016/j.pedhc.2009.09.004
- Broom, A. (2005). The impact of internet use on the disease experience and the doctor patient relationship. *Health: An Interdisciplinary Journal for the Social Study of Health, Illness and Medicine, 9*(3), 319-338.
- Burke, M. L. (1989). Chronic sorrow in mothers of school-age children with myelomeningocele disability (Doctoral dissertation, Boston University). Retrieved from <http://search.proquest.com/mutex.gmu.edu/pqdtft/docview/303665862/previewPDF/13B67909B043566F4C5/4?accountid=14541>
- Burke, M. L., Hainsworth, M. A., Eakes, G. G., & Lingren, C. L. (1992). Current knowledge and research on chronic sorrow: A foundation for inquiry. *Death Studies, 16*(3), 231-245. doi: 10.1080/07481189208252572

- Burke, S. O. (1999). Trajectories and transferability: Building nursing knowledge about chronicity. *Canadian Journal of Nursing Research* 30(4), 243-247.
- Canam, C. (1993). Common adaptive tasks facing parents of children with chronic conditions. *Journal of Advanced Nursing*, 18, 46-53.
- Chien, W.T., Chan, S., Morrissey, J., & Thompson, D. (2005). Effectiveness of a mutual support group for families with patients with schizophrenia. *Journal of Advanced Nursing*, 51(6), 595-608.
- Coffey, J. S. (2006). Parenting a child with chronic illness: A metasynthesis. *Pediatric Nursing*, 32(1), 51-59.
- Coulson, N. S. & Greenwood, N. (2011). Families affected by childhood cancer: An analysis of the provision of social support within online support groups. *Child: Care, Health and Development*, 38(6), 870-877. doi: 10.1111/j.1365-2214.2011.0136x
- Creswell, J. W. (2007). *Qualitative Inquiry and Research Design: Choosing Among Five Approaches*. Thousand Oaks: Sage Publications.
- Croezen, S., Picavet, S. J., Haveman-Nies, A., Verschuren, W. M., De Groot, L., & van't Veer, P. (2012). Do positive or negative experiences of social support relate to current and future health? Results from the Doetinchem cohort study. *BMC Public Health*, 12(65). Retrieved from <http://www.biomedcentral.com/1471-2458/12/65>
- Cutrona, C. E. & Russell, D. (1990). Type of social support and specific stress: Toward a theory of optimal matching. In B. R. Saraason, I. G. Sarason, & G. R. Pierce (Eds.), *Social Support: An Interactional View* (pp. 319-366). New York, NY: Wiley.
- Dabrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mothers and fathers of preschool age children with autism and Down syndrome. *Journal of Intellectual Disability Research*, 54(3), 266-280.
- Damrosch, S. P., & Perry, L. A. (1989). Self-reported adjustment, chronic sorrow, and coping of parents of children with Down syndrome. *Nursing Research*, 38, 25-30.
- Dedoose Version 4.5, web application for managing, analyzing, and presenting qualitative and mixed method research data (2013). Los Angeles, CA: SocioCultural Research Consultants, LLC ([www.dedoose.com](http://www.dedoose.com)).



- Dellve, L., Samuelsson, L., Tallborn, A., Fasth, A., & Hallberg, L. R. (2006). Stress and wellbeing among parents and children with rare diseases: A prospective intervention study. *Journal of Advanced Nursing*, 53(4), 392-402.
- Dolce, M. C., (2011). The internet as a source for health information: Experiences of cancer survivors and caregivers with health providers. *Oncology Nursing Forum*, 38(3), 353-359.
- Eakes, G. G. (1995). Chronic sorrow: The lived experience of parents of chronically mentally ill individuals. *Archives of Psychiatric Nursing*, 9(2), 77-84.
- Eakes, G. G., Burke, M. L., & Hainsworth, M. A. (1998). Middle range theory of chronic sorrow. *The Journal of Nursing Scholarship*, 30(2), 179-184.
- Eastin, M. S. & LaRose, R. (2005). Alt.support: modeling social support online. *Computers in Human Behavior*, 21, 977-992.
- Gage, E. A. & Panagaskis, C. (2011). The devil you know: Parents seeking information online for paediatric cancer. *Sociology of Health and Illness*, 34(3), 444-458. doi: 10.1111/j.1467-9566.2011.01386.x
- Graziosi, E. (2010). Support groups. *The Exceptional Parent*, 40(4), 24.
- Griffin, T., & Kearney, P. M. (2001). Between joy and sorrow: Being a parent of a child with a developmental disability. *Journal of Advanced Nursing*, 35(5), 582-589.
- Gundersen, T. (2011). 'One wants to know what a chromosome is': The internet as a coping resource when adjusting to life parenting a child with a rare disorder. *Sociology of Health and Illness*, 33(1), 81-95. doi: 10.1111/j.1467-9566.2010.01277.x
- Hall, W., & Irvine, V. (2008). E-communication among mothers of infants and toddlers in a community-based cohort: A content analysis. *Journal of Advanced Nursing*, 65(1), 175-183.
- Hainsworth, M. A. (1994). Living with multiple sclerosis: The experience of chronic sorrow. *Journal of Neuroscience Nursing*, 26(4), 237-240.
- Hainsworth, M. A. (1996). Helping spouses with chronic sorrow related to multiple sclerosis. *Journal of Psychosocial Nursing and Mental Health Services*, 34(6), 36-42.

- Han, R., & Belcher, A. E. (2001). Computer-mediated support-group use among parents of children with cancer – an exploratory study. *Computers in Nursing, 19*(1), 27-33.
- Hanson, S. M., Kaakinen, J.R. & Gedaly-Duff, V. (2005). *Family health care nursing: Theory, practice and research*. Philadelphia, PA: F. A. Davis.
- Hatton, D. L., Canam, C., Thorne, S., & Hughes, A. M. (1995). Parents' perceptions of caring for an infant or toddler with diabetes. *Journal of Advanced Nursing, 22*(3), 569-577.
- Hobdell, E. F. (2004). Chronic sorrow and depression in parent of children with neural tube defects. *Journal of Nursing Neuroscience, 36*(4), 82-94.
- Hobdell, E. F., & Deatrck, J. A. (1996). Chronic sorrow: A content analysis of parental differences. *Journal of Genetic Counseling, 5*(2), 57-68.
- Hobdell, E. F., Grant, M. L., Valencia, I., Mare, J., Kothare, S. V., Legido, A., & Khurana, D. S., (2007). Chronic sorrow and coping in families if children with epilepsy. *Journal of Neuroscience Nursing, 39*(2), 76-82.
- Hodges, L. & Dibb, B. (2010). Social comparison within self-help groups: Views of parents of children with Duchenne Muscular Dystrophy. *Journal of Health Psychology, 15*(4), 483-492.
- Hodgkinson, R., & Lester, H. (2002). Stresses and coping strategies of mothers living with a child with cystic fibrosis: Implications for nursing professionals. *Journal of Advanced Nursing, 39*(4), 377-383.
- Hooshmand, M. A. (2010). Comparison of telemedicine to traditional face to face care for children with special health care needs: Analysis of cost, caring and family centered care. (Doctoral dissertation, University of Miami). Retrieved from: <http://search.proquest.com.mutex.gmu.edu/pqdtft/docview/577589679/1431C55155C742C3B8B/2?accountid=14541>
- Hovey, J. (2005). Fathers parenting chronically ill children: Concerns and coping strategies. *Issues in Comprehensive Pediatric Nursing, 28*, 83-95.
- Hovey, J. (2006). Differences in parenting needs of fathers of children with chronic conditions related to family income. *Journal of Child Health Care, 10*(1), 43-54.
- Hunyard, C. (2009). What if anything, is specific about having a rare disorder? Patients' judgments on being ill and rare. *Health Expectations, 12*, 361-370.

- Hymovich, D.P. (1984). Development of the chronicity impact and coping instrument: Parent questionnaire (CICI:PQ). *Nursing Research*, 33, 218-222.
- Institute of Medicine (2011). *The future of nursing: Leading change, advancing health*. Washington, DC: The National Academies Press. Retrieved from: [http://www.nap.edu/openbook.php?record\\_id=12956](http://www.nap.edu/openbook.php?record_id=12956)
- Ireys, H. T., Chernoff, R., DeVet, K. A., & Young, K. (2001). Maternal outcomes of a randomized controlled trial of community-based support program for families of children with a chronic illness. *Archives of Pediatrics and Adolescent Medicine*, 155(7), 771-777.
- Isaksson, A. K., & Ahlstrom, G. (2008). Managing chronic sorrow: Experiences of patients with multiple sclerosis. *The Journal of Neuroscience Nursing*, 40(3), 180-191.
- Isaksson, A. K., Gunnarsson, L. G., & Ahlstrom, G. (2007). The presence and meaning of chronic sorrow in patients with multiple sclerosis. *Journal of Clinical Nursing*, 16(11C), 315-324.
- Issacs, D. & Sewell, J. R. (2003). Children with chronic health conditions. *Medical Journal of Australia*, 1, 235-236.
- Itzhaky, H. & Schwartz, C. (2000). Empowerment of parents of children with disabilities: The effect of community and personal variable. *The Journal of Social Work*, 5(1), 21-36.
- Kamath, B. M., Bauer, R. C., Loomes, K. M., Chao, G., Gerfen, J., Hutchinson, A., Hardikar, W., Hirschfield, G., Jara, P., Krantz, I. D., Lapunzina, P., Leonard, L., Ling, S., Ng, V. L., Hoang, P. L., Piccoli, D. A., & Spinner, N. B. (2012). Notch2 mutations in Alagille syndrome. *Journal of Medical Genetics*, 49, 138-144. doi:10.1136/jmedgenet-2011-100544
- Katz, S. (2002). Gender differences in adapting to a child's chronic illness: A causal model. *Journal of Pediatric Nursing*, 17(4), 257-269.
- Kerr, S. M. & McIntosh, J. B. (2000). Coping when a child has a disability: Exploring the impact of parent-to-parent support. *Child Care Health and Development*, 26(4), 309-321.
- Laskar, J. N., Sogolow, E. D., & Sharim, R. R. (2005). The role of online community for people with rare disease: Content analysis of messages posted on a primary

biliary cirrhosis mailing list. *Journal of Medical Internet Research*, 7(1), e10.doi:  
[10.2196/jmir.7.1.e10](https://doi.org/10.2196/jmir.7.1.e10)

- Law, M., Stewart, D., & King, G. (2001). The perceived effects of parent-led support groups for parents of children with disabilities. *Physical and Occupational Therapy in Pediatrics*, 21(2), 29-48.
- Lazarus, R. S. & Folkman, S. (1984). *Stress, appraisal and coping*. New York, NY: Basic Books.
- Lazarus, R. S. & Folkman, S. (1985). Ways of coping-revised. Retrieved from:  
<http://caps.ucsf.edu/uploads/tools/surveys/pdf/Ways%20of%20coping.pdf>
- Leonard, H., Slack-Smith, L., Phillips, T., Richardson, S., D'Orsogna, L., & Mulroy, S. (2004). How can the internet help parents of children with rare neurologic disorders? *Journal of Child Neurology*, 10(11), 902-907.
- Lichtenstein, B, Laska, M. K., & Clair, J. M. (2002). Chronic sorrow in the HIV positive patient: Issues of race, gender and social support. *AIDS Patient Care and STDS*, 16(1), 27-38.
- Liedstrom, E., Isaksson, A. K., & Ahlstrom, G. (2008). Chronic sorrow in next of kin patients with multiple sclerosis. *Journal of Neuroscience Nursing*, 40(5), 304-311.
- Lincoln, Y. S. & Guba, E. G. (1985). *Naturalistic Inquiry*. Newbury Park: Sage Publications.
- Lindgren, C. L. (1996). Chronic sorrow in persons with Parkinson's and their spouses. *Scholarly Inquiry for Nursing Practice*, 10(4), 351-370.
- Lindgren, C. L., Burke, M. L., Hainsworth, M. A., & Eakes, G. G. (1992). Chronic sorrow: A lifespan concept. *Scholarly Inquiry for Nursing Practice*, 6, 27-40.
- Liu, C., Chao, Y., Huang, C., Wei, F., & Chien, L. (2010). Effectiveness of applying empowerment strategies when establishing a support group for parents of preterm infants. *Journal of Clinical Nursing*, 19, 1729-1737. doi: 10.1111/j1365-2702.2009.03082x
- Lowes, L., & Prowse, M. A. (2001). Standing outside the interview process? The illustrations of objectivity in phenomenological data generation. *International Journal of Advanced Nursing*, 31, 219-255.

- Mallow, G., E., & Bechtal, G. A. (1999). Chronic sorrow: The experience of parents with children who are developmentally disabled. *Journal of Psychosocial Nursing*, 37, 31-35.
- Margalit, M. & Raskind, M. H. (2009). Mothers of children with LD and ADHD: Empowerment through online communication. *Journal of Special Education Technology*, 24(1), 39-45.
- Mason, M. (2010). Sample size and saturation in PhD studies using qualitative interviews. *Forum for Qualitative Social Research*, 11(3). Online. Retrieved from: <http://www.qualitative-research.net/index.php/fqs/article/view/1428/3027>
- Maxwell, J. A. (2013). *Qualitative research design: An interactive approach*. Los Angeles, CA: Sage Publications.
- McCubbin, H. I., McCubbin, M. A., Nevin, R., & Cauble, E. (1981). Coping-health inventory for parents (CHIP). In H. I. McCubbin, A. I. Thompson & M. A. McCubbin (Eds; 1996). *Family assessment: Resiliency coping and adaptation-inventories for research and practice*. Madison, WI: University of Wisconsin System.
- McDaniell, R., Warthen, D. M., Sanchez-Lara, P. A., Pai, A., Krantz, I. D., Piccoli, D. A., & Spinner, N. B. (2006). Notch2 mutations cause Alagille syndrome, a heterogeneous disorder of the notch signaling pathway. *American Journal of Human Genetics*, 79, 169-173. Doi: 10.1086/505332
- McElhinney, D. B., Kranz, I. D., Bason, L., Piccoli, D. A., Emerick, K. M., Spinner, N. B., & Goldmunz, E. (2002). Analysis of cardiovascular phenotype and genotype-phenotype correlation in individuals with a JAG1 mutation AND/OR Alagille syndrome. *Circulation*, 106, 2567-2574. doi: 10.1161/01.CIR.0000037221.45902.69
- McGrath, P., & Chesler, M. (2004). Fathers' perspective on the treatment for pediatric hematology: Extending the findings. *Issues in Comprehensive Pediatric Nursing*, 27, 39-61.
- Meleski, D. (2002). Families with chronically ill children: A literature review examines approaches to helping them cope. *American Journal of Nursing*, 102(5), 47-54.
- Morahan-Martin, J. M. (2004). How internet users find, evaluate, and use online health information: A cross cultural review. *Cyberpsychology & Behavior*, 7(5), 497-510.

- Mosby's Medical Dictionary (2009). Retrieved from <http://medical-dictionary.thefreedictionary.com/psychosocial>
- Munhall, P. L. (2012). *Nursing research: A qualitative perspective*. Sudbury, MA: Jones and Bartlett Learning.
- National Digestive Diseases Information Clearing House (2009). Alagille syndrome. Retrieved from <http://digestive.niddk.nih.gov/ddiseases/pubs/alagille/index.aspx>
- National Institutes of Health (2012). Rare Disease Day information fact sheet. Retrieved from <http://rarediseases.info.nih.gov/RareDiseaseDay.aspx>
- National Institutes of Health (2011, Feb). Rare Diseases Network fact sheet. Retrieved from <http://report.nih.gov/NIHfactsheets/ViewFactSheet.aspx?csid=126&key=R#R>
- National Institutes of Health, Offices of Rare Disease Research (n.d.). Rare diseases. Retrieved from <http://rarediseases.info.nih.gov/Resources.aspx?PageID=32>
- Nordfeldt, S., Angarne-Lindberg, T., Nordwell, M., & Krevers, B. (2013). Parents of children with type 1 diabetes – their views on information and communication needs and internet use. A qualitative study. *Parents of Adolescents with Type 1 Diabetes*, 8(4), 1-8. doi: 10.1371/journal.pone.0062096
- Northington, L. (2000). Chronic sorrow in caregivers of school age children with sickle cell disease: A grounded theory approach. *Issues in Comprehensive Pediatric Nursing*, 23, 141-154.
- Olshansky, S. (1962). Chronic sorrow: A response to having a mentally defective child. *Social Casework*, 43, 190-193.
- Patterson, J. M. (2002). Integrating family resilience and family stress theory. *Journal of Marriage and Family*, 64(2), 349-360.
- Perrin, E., Lewkowicz, C., & Young, M. (2000). Shared vision: Concordance among fathers, mothers, and pediatricians about unmet needs of children with chronic health conditions. *Pediatrics*, 105(1), 277-285.
- Porter, A. & Edirippulinge, S. (2007). Parents of deaf children seeking hearing loss-related information on the internet: The Australian experience. *Journal of Deaf Studies and Deaf Education*, 12(4), 518-529.

- Pew Internet and American Life Project (2007). *How gender influences health searches*. Retrieved from <http://www.pewinternet.org/Commentary/2007/August/How-Gender-Influences-Health-Searches.aspx>
- Pew Internet and American Life Project (2011,a). *Search and email still the most popular online activities*. Retrieved from <http://pewresearch.org/pubs/2079/-email-internet-search>
- Pew Internet and American Life Project (2011,b). *The social life of health information, 2011*. Retrieved from <http://www.pewinternet.org/Reports/2011/Social-Life-of-Health-Info.aspx>
- Pew Internet and American Life Project (2012). *Demographics of internet users*. Retrieved from [http://www.pewinternet.org/Static-Pages/Trend-Data-\(Adults\)/Whos-Online.aspx](http://www.pewinternet.org/Static-Pages/Trend-Data-(Adults)/Whos-Online.aspx)
- Pew Internet and American Life Project (2012). *Family Caregivers turn to the internet for information and guidance*. Retrieved from <http://pewresearch.org/pubs/2304/caregivers-internet-online-health-care-disability-long-term-information-treatments-medical>
- Pew Internet and American Life Project (2013). *Demographics of internet users*. Retrieved from [http://www.pewinternet.org/Static-Pages/Trend-Data-\(Adults\)/Whos-Online.aspx](http://www.pewinternet.org/Static-Pages/Trend-Data-(Adults)/Whos-Online.aspx)
- Pew Internet and American Life Project (2013). *72% of online adults are social networking site users*. Retrieved from [http://www.pewinternet.org/~media/Files/Reports/2013/PIP\\_Social\\_networking\\_sites\\_update\\_PDF.pdf](http://www.pewinternet.org/~media/Files/Reports/2013/PIP_Social_networking_sites_update_PDF.pdf)
- Resendez, M., Quisti, R., & Matashazi, D. (2000). A longitudinal analysis of family empowerment and client outcomes. *Journal of Child and Family Studies*, 9(4), 449-460.
- Roche, M. I. & Skinner, D. (2009). How parents search, interpret, and evaluate genetic information obtained from the internet. *Journal of Genetic Counseling*, 18, 199-129.
- Rose, P., Beeby, J., & Parker, D. (1995), Academic rigour in the lived experience of researchers using phenomenological methods in nursing. *Journal of Advanced Nursing*, 21, 1123-1129.
- Rosenberg, C. J. (1998). Faculty-student mentoring: A father's chronic sorrow: A daughter's perspective. *Journal of Holistic Nursing*, 16(3), 399-406.

doi: 10.1177/089801019801600308

- Schaffer, R., Kuczynski, K., & Skinner, D. (2008). Producing genetic knowledge and citizenship through the internet: Mothers, pediatric genetics, and cybermedicine. *Sociology of Health and Illness*, 30(1), 145-159.
- Scharer, K. (2005). Internet social support for parents: The state of the science. *Journal of Child and Adolescent Psychiatric Nursing*, 18(1), 26-33.
- Scornaienchi, J. M. (2003). Chronic sorrow: One mother's experience with two children with lissencephaly. *Journal of Pediatric Health Care*, 17, 290-294.
- Semere, W., Karamanoukian, H. L., Levitt, M., Edwards, T., Murero, M., D'Acona, G., Donias, H. W., Glick, P. L. (2003). A pediatric surgery study: Parent usage of the internet for medical information. *Journal of Pediatric Surgery*, 38(4), 560-564.
- Skinner, D. & Schaffer, R. (2006). Families and genetic diagnoses in the genomic and internet age. *Infants and Young Children*, 19(1), 16-24.
- Smith, C. S. (2009). Substance abuse, chronic sorrow and mother loss: Relapse triggers among female victims of child abuse. *Journal of Pediatric Nursing*, 24(5), 401-412.
- Smith, K. & Schaefer, A. (2012). *Who cares for sick kids? Parents' access to shared time to care for a sick child* (Spring Issue Brief No. 51). Retrieved from the Carsey Institute website: <http://www.carseyinstitute.unh.edu/publications/IB-Smith-Paid-Sick-Leave-2012.pdf>
- Solomon, M., Pistrang, M., & Barker, C. (2001). The benefits of mutual support groups for parents of children with disabilities. *American Journal of Community Psychology*, 29(10), 113-130.
- Spinner, N. B., Colliton, R. P., Crosnier, C., Krantz, I. D., Hadchouel, M., & Meunier-Rotival, M. (2001). Jagged mutations in Alagille syndrome. *Human Mutation*, 17(1), 18-33.
- Stepney, C. S., Kane, K., & Bruzzese, J. (2011). My child is diagnosed with asthma, now what?: Motivating parents to help their children control asthma. *The Journal of School Nursing*, 27(5), 340-347.
- Streubert, H. J. & Carpenter, D. R. (2011). *Qualitative research in nursing: Advancing the humanistic imperative*. Philadelphia, PA: Wolters Kluwer.



- Sullivan, C. (2008). Cybersupport: Empowering asthma caregivers. *Pediatric Nursing*, 34(3),217-224.
- Svavarsdottir, E. K., & Rayens, M. K. (2003). American and Icelandic Parents' Perceptions of the Health Status of their Young Children with Chronic Asthma. *Journal of Nursing Scholarship*, 35(4), 351-358.
- Swallow, V. M. & Jacoby, A. (2001). Mother's evolving relationship with doctors and nurses during the chronic childhood illness trajectory. *Journal of Advanced Nursing*, 36(6), 755-764.
- Teel, C. S. (1991). Chronic sorrow: Analysis of the concept. *Journal of Advanced Nursing*, 16, 1311-1319.
- Timmermans, S. & Oh, H. (2010). The continued social transformation of the medical profession. *The Journal of Health and Social Behavior*, 51(1), S94-106.
- Tozzi, A. E., Mingarelli, R., Agricola, E., Gonfiantini, M., Pandolfi, E., Carloni, E., Gesualdo, F., & Dallapiccola, B. (2013). The internet user profile of Italian families of patients with rare diseases: A web survey. *Orphanet Journal of Rare Diseases*, 8(76), 1-8.
- Trollvik, A. & Severinsson, E. (2005). Influence on asthma education program on parents of children suffering from asthma. *Nursing and Health Sciences*, 7, 157-163.
- Tucker, C. M., Butler, A. M., Loyuk, I. S., Desmond, F. F., & Surrency, S. L. (2009). Predictors of a health-promoting lifestyle and behaviors among low-income African American mothers and White mothers of chronically ill children. *Journal of the National Medical Association*, 101(2), 103-110.
- U.S. Department of Health and Human Services (2012).*E-Health overview*. Retrieved from <http://www.health.gov/communication/ehealth/>
- van Manen, M. (1990). *Researching the lived experience: Human science for action sensitive pedagogy*. Albany, NY: State University of New York Press
- Vickers, M. H. (2005). Bounded grief at work: working and caring for children with chronic illnesses. *Illness, Crisis and Loss*, 13(3), 201-218.
- Walsh, F. (2006). *Strengthening family resilience*. New York, NY: Guildford

- Wang, H. H., Wu, S. Z., & Liu, Y. Y. (2003). Association between social support and health outcomes: A meta-analysis. *Kaohsiung Journal of Medical Sciences*, 19(7), 345-351.
- Weiten, W., Dunn, D. S., & Hammer, E. Y. (2011). *Psychology applied to modern life: Adjustments in the 21<sup>st</sup> century*. Belmont, CA: Wadsworth.
- Yi, Y. J., Stvilia, B., & Mon, L. (2011). Cultural influences on seeking quality health information: An exploratory study of the Korean community. *Library and Information Science Research*, 1-22.
- Ziebland, S. (2004). The importance of being expert: the quest for cancer information on the internet. *Social Science and Medicine*, 59, 1783-1793.

## **BIOGRAPHY**

Adriana Drake Glenn was born in Cleveland, Ohio. She received her Bachelor of Science in Nursing from Boston University. She earned her Master of Nursing from the University of California, Los Angeles concentrating in Primary Ambulatory Care/Family Nurse Practitioner and her Master of Arts concentrating in Education Administration with a focus on Education Leadership from California State University, Los Angeles. She is currently employed as adjunct faculty member at George Mason and Marymount Universities. In addition, she maintains her practice via employment as a nurse practitioner for the Virginia Department of Health, Alexandria, Virginia. Mrs. Glenn's clinical experiences include cardiac critical care, providing health care in educational settings (K-12 public schools, higher education institutions), and working in a variety of community and public health environments including urgent care, occupational/employee health, community clinics, and public health clinics/departments. Mrs. Glenn also has extensive experience in teaching in a variety of academic environments including: clinical instruction, precepting, classroom lecturer and lab instructor at the community college, undergraduate, and graduate levels.

Mrs. Glenn developed her research interest in online communication/e-health communication, rare diseases and parental coping/social supports in response to her experiences with her daughter Avery who had Alagille syndrome. Mrs. Glenn is an active member of the Alagille Syndrome Alliance. Upon completion of her doctorate, she plans to seek faculty appointment at a university.