THE LIVED EXPERIENCE OF FATHERS CARING FOR THEIR CHILD WITH CYSTIC FIBROSIS

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DEDICATION

For my husband and son, Felix and Will, thank you for never-ending support and love.

To my parents, Jack and Lucille Husung, thank you for always believing in me.

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ABSTRACT

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Cystic fibrosis (CF) is a life-shortening genetic disease with many treatment requirements that necessitate the participation of a caregiver, especially if the patient is a child. Most studies of the quality of life of caregivers of children with CF have focused on the mental health of mothers, reflecting a biased underlying assumption that mothers are the primary caregivers. The aim of this study was to explore the experience of fathers caring for a child with CF. Interviews were conducted with 20 fathers of children with CF. Interviews were semi-structured using Husserl's descriptive phenomenology. The six themes emerged from the interviews were: feeling overwhelmed, feeling isolated, experiencing altered family dynamics, actively seeking resources, experiencing financial strain, and feeling hope. Fathers of children with CF reported distressing experiences in connection with their child's diagnosis of CF and during the course of their child's disease, but they also a had strong feelings of hope for the future.

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CHAPTER I

INTRODUCTION

Cystic fibrosis (CF) is the most common life-shortening genetic disease of the Caucasian population affecting 70,000–100,000 people worldwide (Cystic Fibrosis Worldwide, 2013). Cystic fibrosis is a multi-systemic disease that requires intensive treatments including chest physiotherapy and multiple inhaled and oral medications that require administration several times per day. Many of these therapies demand active participation of a caregiver, especially if the patient is a child. The treatment burden is high for both the child and his or her caregiver. The potential implications of this treatment burden include decreased quality of life for the caregiver.

The role of the father in caring for seriously ill children is often overlooked due to the mother more often being the primary caretaker. In a seminal research study, Gayton, Friedman, Tavormina, and Tucker (1977) reported that fathers of CF children experienced higher rates of emotional disturbance compared to the general population and even higher rates than mothers of CF patients. More recently, a study by Glasscoe, Lancaster, Smyth, and Hill (2007) corroborated this finding, reporting that fathers of newly diagnosed CF patients experienced more dysphoria compared to non-CF fathers. On the other hand, a study conducted by Driscoll, Montag-Leifling, Acton, and Modi (2009) found that while depression rates were elevated in CF caregivers, depression rates among male and female primary caregivers did not differ. The majority of studies that

have been conducted assessing the father as caregiver have been quantitative studies, and male participation has been historically low compared to female participation (approximately 4:1).

In Ireland, one published qualitative study on the experience of fathers caring for their preschool children with CF was found (Hayes & Savage, 2008). Clinical care guidelines set by the European Cystic Fibrosis Trust (Colombo & Littlewood, 2011) and the U.S. Cystic Fibrosis Foundation (Lahiri et al., 2016; Mogayzel et al., 2013) show differences in the care that young children receive in Europe versus the United States, which could have a geographic or cultural effect on how a father perceives the experience of caring for his child with CF. To the researcher's knowledge, there are no published qualitative studies on the lived experience of fathers caring for their children with CF in the United States. The fathers of children with CF likely have many different experiences while caring for their children, which in turn can affect the whole family unit, but this has not been adequately assessed. Thus, this research examined the lived experience of fathers in the United States caring for children with CF.

Statement of Purpose

The purpose of this qualitative research study is to describe the lived experience of fathers caring for a child with CF in the United States. Information on the role and function of the father of CF children within the family paradigm is limited. To date, the research has focused on mothers of children with CF, and a significant gap exists in the literature related to the role of the father within the family unit.

Research Question

The overarching research question that will be addressed in this study is as follows: What is the lived experience of fathers caring for a child with CF? Additional questions and probes will be used to guide the interviews. The specific aims of this study include:

- To describe common meanings and shared experiences of fathers caring for their child/children with CF.
- To explore factors that influences the experience of fathers of a child with CF, such as the age of the child and the time since CF diagnosis.

Rationale for the Study

Background of the Study

Much of the research on caring for seriously ill children focuses on mothers because mothers are often the primary caregiver. In an original research study, Gayton et al. (1977) studied 43 mothers and 29 fathers of CF children and found that 40% of fathers and 24% of mothers experienced emotional disturbances. The prevalence of emotional disturbances experienced in the control group of non-CF mothers was similar to that experienced by the mothers of CF patients, but the control group of non-CF fathers experienced emotional disturbances at a lower rate, suggesting that fathers of CF patients were at increased risk in this study. Gayton et al. linked the financial stress imposed by cystic fibrosis with depression suffered by fathers of children with CF. Gayton et al. stated that CF is a costly disease, and the economic impact likely affects the depression

rates of fathers. On the other hand, Driscoll et al. (2009) found that while depression and anxiety rates were elevated in CF caregivers and concordant in participating caregiver dyads, rates of depression and anxiety among male and female caregivers were similar. Family stress and unemployment are important risk factors for depression in families caring for children with CF (Driscoll et al., 2009). Economic factors are considerable for fathers of CF patients, but other risk factors related to depression and anxiety need to be more adequately assessed in the paternal role.

Glasscoe et al. (2007) examined the prevalence of dysphoria in parents of newly diagnosed children with CF compared to matched controls (parents of healthy children). They reported that parents of CF children $\leq$ 9 months of age were at higher risk of dysphoria; the average values for relative risk (RR_{MH}) for fathers and mothers under baseline conditions were 3.51 and 2.1, respectively. In addition, the risk of dysphoria for this group of parents remained elevated during follow-up, with the average values for fathers and mothers' RR_{MH} being 2.6 and 2.26, respectively.

When assessing the literature of CF caregivers, most studies conducted quantitative assessments for depression and anxiety due the burden of the disease. It is reasonable to believe that as a caregiver experiences increased levels of depression or anxiety, his or her quality of life will also suffer. Depression and anxiety were found to be common variables analyzed when assessing quality of life in CF caregivers (Besier et al., 2011; Besier & Goldbeck, 2011; Driscoll et al., 2009; Glasscoe et al., 2007; Quittner et al., 2014; Smith, Modi, Quittner, & Wood, 2010).

The depression rate of the general public has been reported at a rate of 8% (American Psychological Association [APA], 2010). In an international multi-center study, it was found that 37% of mothers (n = 3,127) and 31% of fathers (n = 975) of children with CF reported elevated scores of depression, suggesting depression (Quittner et al., 2014). Driscoll et al. (2009) reported the anxiety rates in female (n = 100) and male (n = 22) caregivers of children with CF, although the female-to-male ratio was 4:1 in the sample. Driscoll et al. found that 43% of male and 51% of female caregivers had anxiety. APA (2010) reported the rate of anxiety in the general population is approximately 21%, meaning that Driscoll et al.'s (2009) findings indicate that CF caregivers have as much as a two-fold rate of anxiety as the general population.

Hayes and Savage (2008) performed the only qualitative study found in the literature on the experience of fathers caring for their children with CF. The study, conducted in Ireland, examined a homogenous group of eight patients. All fathers were married, had preschool male children, and had wives who stayed home with the child/children.

Significance to Nursing Practice

Nurses are the cornerstone of care at CF centers across the United States and are usually the first contact for the families of children with CF. Nurses provide CF education and care, and they are the first line of resource when a patient or family needs a referral. Decreased caregiver quality of life affects the entire family, as well as the health of the ill child if the caregiver cannot function at an optimal capacity to care for the child

with CF. Gordon (2009) reported that chronic stress in the family of a chronically ill child could have an adverse effect on the health of the child and family.

Many families deal with the disease of CF differently across the lifespan. The disease can bring families closer together, or it can have devastating consequences. The perspective of the father within the family unit, whether or not he is the primary caregiver or plays a lesser role, has not been explored well. Concern with the mental health of the child and his or her family has recently come to the forefront in care of patients with CF and is now recognized as a main goal in CF care.

Underlying Assumptions

Few researchers enter a field of research without underlying assumptions or preconceived beliefs about the research topic. Polit and Beck (2012) stated that assumptions are beliefs that have no proof. The assumptions leading into this research include:

- The majority of mothers do most of the work when it comes to caring for their child with CF.
- Fathers play an integral role in the life of the family unit, and their ability to cope and care for their child with CF affects the entire family.
- Fathers would like to be more involved in the care of their child/children with CF.
- Psychological interventions/follow-ups for families would have a positive impact on their quality of life.

- Psychological intervention for the family would positively impact the child's adherence to treatments and ultimately the outcomes with CF.

Philosophical Framework

The philosophical framework of this study will be Husserl's (1970) descriptive phenomenology. Edmund Husserl is the philosopher affiliated with descriptive phenomenology (Audi, 2015). Husserl, a German philosopher, is credited with being the father of phenomenology in the late 1800s to early 1900s (Audi, 2015). Husserl started his academic career as a mathematician but soon began to feel that science attempted to marginalize humans, and he began his search for the meaning of life in the experiences of people, which is referred to as intentionality and intuition in his works (Audi, 2015). Husserl felt that people could not exist without the experiences of their past and their present, and that their experiences shape who they are as people (Audi, 2015). Husserl believed that everyone possesses a transcendental ego that plays a key role in the way people respond to other people, to the world that shapes the conscious, and to what he referred to as the living present (Audi, 2015).

While descriptive phenomenology is a multifaceted methodology and philosophy, three basic underpinnings of Husserl's philosophy will be used to guide this research study. The first of these is the underpinning that a one-on-one interaction with participants creates a reflection of reality (Husserl, 1970). The second underpinning used to guide the study will be the use of transcendental subjectivity. Transcendental subjectivity, or bracketing, is when the researcher sets aside his or her own beliefs and

reality and describes a phenomenon in a purist way (Husserl, 2001). The third underpinning that will be used to guide this study is the belief that each person has the ability to influence his or her own environment (Husserl, 2001). The experience may be different (or not) for each participant. The goal of phenomenology is to describe the life experiences of people in an analytical way that will advance the science. This corresponds with the goal of this study, which is to explore and, thus, have a better understanding of the lived experiences of fathers who have children with cystic fibrosis.

Summary

Many advances have been made in the care of patients with CF. These advances have led to an increased life span for patients. While this is extraordinary, CF remains a disease with high treatment burden, and with every advancement comes increased burden for both the patient and caregiver. While the mother is often the primary caregiver, the role of the father is often overlooked, and this has been an oversight on the part of healthcare providers. Fathers, like mothers, are an important part of the family unit, yet their experience of caring for their children with CF has not been adequately explored across different age spans and sexes of children with CF in the United States. The fact that little research has been conducted about fathers, especially related to their experience caring for their children with CF, is concerning. Understanding fathers' experiences can assist nurses in guiding families in caring for themselves and ultimately caring for their children with CF in the best possible way.

CHAPTER II

REVIEW OF LITERATURE

A Paper Submitted for Publication in the

Pediatric Nursing Journal

Quality of Life in Cystic Fibrosis Caregivers: An Integrated Literature Review

Abstract

Cystic fibrosis (CF) is a life-shortening genetic disease with many treatment requirements that necessitate the participation of a caregiver, especially if the patient is a child. We performed an integrated literature review of original research evaluating the quality of life for caregivers of patients with CF. We performed a search using the terms *quality of life*, *cystic fibrosis*, and *caregivers* in the MEDLINE, CINAHL, Child Development & Adolescent Studies, PsycARTICLES, and Psychology & Behavioral Sciences Collection databases from 2007 to 2017. Nine articles were included. They showed that caregiver quality of life is affected by caring for a child with CF, as evidenced by high rates of anxiety and depression. Anxiety and depression in caregivers affects adherence to the child's treatment regimen, causing detrimental effects on the health of the child. Screening for depression and anxiety with referral for treatment when necessary are needed to optimize family health.

Keywords

quality of life, parents, anxiety, depression, parental health, cystic fibrosis, mental health

Highlights

- Cystic fibrosis (CF) has numerous treatment burdens for caregivers.
- Rates of anxiety and depression are two to three times higher for caregivers of patients with CF than in the general population.
- Depressed parents are less likely to adhere to their child's treatment regimen.
- Few CF families report having received mental health screening or treatment.

Cystic fibrosis (CF) is the most common life-shortening genetic disease in the White population; affecting 70,000 people worldwide (Cystic Fibrosis Worldwide, 2018). While many healthcare professionals think of CF primarily as a lung disease, historically CF first presented as children dying of malnutrition related to pancreatic insufficiency and malabsorption, which affect up to 90% of individuals with CF. The disease was first noted in modern medicine by pathologist Dorothy Anderson, who described the condition as "cystic fibrosis of the pancreas," which she discovered while performing an autopsy on a child (National Institutes of Health [NIH], 2018). At that time, children with CF did not live past infancy. Anderson's discovery and the subsequent treatment of the pancreatic insufficiency with pancreatic enzymes was a major advancement that contributed to the survival of patients past the first year of life. Major improvements in modern medicine over the next 30 to 40 years advanced the life expectancy of people with CF in the United States from infancy to a now median age of 47 years (Cystic Fibrosis Foundation [CFF], 2016).

CF is caused by pathogenic mutations of the CF transmembrane conductance regulator, which causes an abnormal secretion of chloride and other anions. This abnormal secretion of anions leads to thick, tenacious secretions and exocrine dysfunction, resulting in a multisystemic disease (Brennan & Schrijver, 2016). The CF gene, along with its major disease-causing mutation, was a relatively recent discovery that occurred in 1989. Despite these advances in treatments, CF is a progressive disease and patients still continue to lose 1 to 3 percentage points of lung function per year (Liou et al., 2010). While CF remains a multisystemic disease, today, 90% of patients with CF die from pulmonary disease or a complication related to treatment (CFF, 2016). Prevention of pulmonary exacerbations and maintenance of general health is the goal of treatment.

As previously mentioned, many therapeutic options were developed early on for children with CF, and these have intensified over the past 30 years. The culmination of these therapeutic developments has improved both the quality of life and the life expectancy of patients with CF. These improvements, however, have come at a cost to both patients and caregivers since treatments needed to sustain health are time intensive. Examples of such treatments include chest physiotherapy, which can take up to 1 hour or more, and medication administration, which includes multiple daily doses of inhaled and oral medications. Many CF therapies require the active participation of a caregiver, especially if the patient is a child.

Such burdensome treatment requirements can potentially decrease caregiver quality of life and can contribute to health problems for the caregiver. Caregivers of patients with CF frequently report depression related to their loved one's diagnosis (Barker & Quittner, 2016; Quittner et al., 2016). Anxiety is often a comorbidity with depression, but studies published recently have found that the prevalence of anxiety is higher than that of depression in caregivers of patients with CF (Driscoll, Montag-Leifling, Acton, & Modi, 2009). Relentless depression and anxiety related to a life-threatening illness decreases the quality of life of these caregivers, which can affect the entire family, as well as the health of the ill child, if the caregiver cannot function at optimal capacity (Gordon, 2009).

Purpose

The purpose of this integrated review is to provide nurses with a synthesis of the scholarly literature published from 2007 to 2017 that is related to the quality of life of parental caregivers for patients with CF. It is important to understand that CF care is provided at CF Centers of Excellence across the United States and Canada. These centers take a multidisciplinary approach to the care of patients with CF, with nurses at the center of patient care responsibility.

The care of patients with CF is an excellent example of family-centered nursing care and one that can be improved upon. In the past, patients were dying before they reached adulthood, and nurses were focused on the illness and medication aspects of the disease. While this focus will always remain paramount to some extent, nurses now more

than ever can incorporate the psychosocial aspects of the entire family into their treatment plan. This focus has become so clear to the CFF that they issued guidance directing that patients with CF and their families be screened for psychosocial illnesses yearly (Quittner et al., 2016). Nurses are often the first point of contact for all patients with CF and can make the difference in the lives of these families. The findings from this review demonstrate how caregivers of patients with CF are affected and highlight interventions that have the potential to improve the health and well-being of caregivers, the individuals with CF, and their families.

Definition of Caregiver Quality of Life

Quality of life is a multidimensional concept that can differ from person to person but in general reflects an individual's ability to enjoy everyday life events. The concept of quality of life has long been an important one, as evidenced by the World Health Organization's attempt to start defining quality of life in 1948. While the constructs are widely debated, most agree that quality of life includes aspects of physical function, mental health, and the ability to engage in social interactions (Centers for Disease Control and Prevention [CDC], 2016).

Methods

A search of the MEDLINE (National Library of Medicine), CINAHL (EBSCO), Child Development & Adolescent Studies (EBSCO), PsycARTICLES (American Psychological Association), and Psychology & Behavioral Sciences Collection (EBSCO) databases was conducted by using the search terms *quality of life*, *cystic fibrosis*, and

caregivers. The date of publication limit placed on the search was from 2007 to 2017. The original search revealed 49 articles. We reviewed the articles and retained those that summarized original research evaluating the quality of life of caregivers. Inclusion criteria included studies of parental caregivers of children with CF published in English. Exclusion criteria included (a) review articles, opinion articles, and clinical updates or original research articles pertaining to the quality of life of caregivers for CF and any other disease process; (b) articles that assessed quality of life as a secondary endpoint while evaluating another primary endpoint; and (c) articles that dealt with family caregivers (other than parental), hired caregivers, professional caregivers, and lay-caregivers. Of the original 49 articles, nine were included in the final review.

Results

This literature review found that parental caregiver quality of life is greatly impacted when caring for a child with CF. Parental caregivers reported experiencing depression, anxiety, coping issues, and financial stress related to caring for their children with CF. Parental caregivers of CF patients also were found to report on quality of life satisfaction as well as adherence to CF treatment in the literature review. Those findings are addressed in more detail below.

In addition, a summary of the details of the final nine studies, including author, publication date, sample, and findings are listed in Table 2.1. Various tools were used to measure quality of life, and many of the articles used multiple tools. Some tools measured quality of life directly, such as by using the Caregiver Quality of Life-Cystic

Fibrosis (CQOL-CF) scale, or indirectly by screening for depression symptoms with the Center for Epidemiological Studies Depression Scale (CES-D). Table 2.2 reviews the different tools used in this review of literature as well as the reliability and validity data available on each tool, as explicitly stated in each article.

Depression

The belief exists that as a caregiver experiences increased levels of depression or anxiety, quality of life will be impacted. Depression is a common variable when assessing the quality of life of caregivers. A common tool used to assess quality of life in this review was the CES-D. Caregivers for individuals with CF were found to have higher rates of depression than the general public, which is reported at 8% (American Psychological Association [APA], 2018). In a study of 162 caregivers of patients with CF, Besier and Goldbeck (2011) found that 26.4% had elevated CES-D scores, suggesting clinical depression. In a larger, multicenter study, Besier et al. (2011) corroborated the earlier findings, finding depression rates in the 650 caregivers studied to be 28% when measured by using the CES-D. The largest study was an international, multicenter study by Quittner et al. (2014) using the CES-D to evaluate 4,102 caregivers, which found depression rates of 37% in mothers (n = 3,127) and 31% in fathers (n = 975). Two other studies examined depression in mother/female caregivers as compared to father/male caregivers. In these studies, like the Quittner et al. study, the participation rate was significantly higher for mothers than fathers (Driscoll et al., 2009; Smith, Modi, Quittner & Wood, 2010).

In the study by Driscoll et al. (2009), which included a sample of 122 caregivers, the women (n = 100) reported depression rates ranging from 20% to 28%, while the men (n = 22) reported rates ranging from 14% to 31%. Comparatively, Smith et al. (2010) reported depression rates in the parents of 39 children with CF as 35% for mothers (n = 21) and 23% for fathers (n = 13). Smith et al. also found that 32% of the children studied had at least one parent with depression. Driscoll et al. (2009) also reported that depression symptoms in women were positively correlated with depression symptoms in men in families where both parents participated as caregivers. If one parent was depressed, the other was also more likely to be depressed. As depression increased in caregivers, their quality of life decreased (Driscoll et al., 2009); however, few caregivers were undergoing psychotherapy or receiving pharmacotherapy for their depression (Besier & Goldbeck, 2011). In a study by Neri, Luicidi, Catastini and Colombo (2016) assessing caregiver burden, 34% of the sample reported scores suggestive of depression. Depression appears to be a significant health-related problem in caregivers.

Anxiety

In a study of 162 parents of children with CF, Besier and Goldbeck (2011) found that 37.7% of the parents had anxiety. A larger cohort study of 650 parents of children with CF (n = 245 cases in which both parents participated; n = 478 cases in which only one parent participated, with n = 73 fathers, n = 405 mothers) supported this finding, with 37.2% of participants having anxiety (Besier et al., 2011). An international, multicenter study found that 48% of mothers (n = 3,127) and 36% of fathers (n = 975) reported

clinical levels of anxiety (Quittner et al., 2014). Driscoll et al. (2009) also reported anxiety in female and male caregivers, although the female-to-male ratio was 4:1 in the sample. Low male participation has been the norm in most CF studies. Driscoll et al. (2009) reported that 43% of male and 51% of female caregivers had anxiety. APA (2018) reported the rate of anxiety in the general population as approximately 21%, indicating that the rate of anxiety found by Driscoll et al. (2009) among caregivers was more than twice that of the general population. Besier and Goldbeck (2011) found a correlation between anxiety in the caregiver and in the patient, yet they found low utilization of mental health services in the caregiver population. High rates of anxiety are a major health concern for both caregivers and patients.

Quality of Life and Life Satisfaction

Studies of caregivers for people with CF have linked anxiety and depression to a decrease in quality of life (Neri et al., 2016; Smith et al., 2010; Driscoll et al., 2009). Other studies have directly measured the quality of life of the caregiver with a specific instrument, namely the CQOL-CF. The CQOL-CF tools measure physical, emotional, family, and social function, as well as areas related to finances and spiritual beliefs (Boling, Macrina, & Clancy, 2003). Driscoll et al. (2009) assessed caregiver quality of life using the CQOL-CF instrument on men and women and found similar scores of 97.74 for women and 100.65 for men. Driscoll et al. (2009) also attempted to link quality of life with depression and anxiety in their study. Driscoll et al. (2009) found that quality of life in women was attributed to better lung function in the child, as evidenced by fewer

depressive and anxiety scores for women whose children had higher lung function. Interestingly, there was a correlation in depression and anxiety scores for men and women who were married partners (Driscoll et al., 2009).

By contrast, a study by Wallenwein, Schwarz, and Goldbeck (2017) evaluated single parents of children with CF ($n = 260$) and found that they had a lower quality of life ($M = 54.89$; $SD = 12.44$) than did partnered parents of children with CF ($M = 61.55$; $SD = 15.43$; $t\ (258) = -2.581$; $p = .010$). Wallenwein et al. stated that their findings were independent of the age of the child with CF and the employment status of the parent.

Caregiver burden and vocational participation were assessed by Neri et al. (2016). In addition to corroborating the findings on caregiver depression (34%), this study found a correlation between increased caregiver burden and the need to make changes or leave a career to care for a child with CF. Although not specifically evaluating single or partnered parents, the findings of Neri et al. appear to refute the findings of Wallenwein et al. (2017). In Neri et al.'s study, caregivers in the higher Caregiver Burden Global Strain Index (CB-GSI) quintiles had statistically significant outcomes for presenteeism (working while sick), sick leave, family leave, and reduced work hours/day ($p < .01$). Importantly, as these studies indicate, quality of life, caregiver burden, and vocational limitations are all substantially affected by caregivers of patients with CF.

Coping

A few of the studies reviewed examined various coping mechanisms employed by caregivers. Wong and Heriot (2008) correlated caregiver coping strategies to quality of

life. Wong and Heriot found that having a strong social support system was associated with less negative health outcomes in caregivers; however, negative coping strategies such as self-blame and disengagement from the disease were associated with adjustment issues in both the parent and the child. Having expectations of positive outcomes in the child's future was associated with parent and child well-being, while the converse also was true (Wong & Heriot, 2008). Besier et al. (2011) found that caregivers had increased satisfaction with family life compared to the general population, although, in contrast, they had little leisure time, less ability to relax, and less satisfaction with their jobs and partners. Mindset and strong social support were important factors influencing parents' coping abilities (Wong & Harriet, 2008; Bessier et al., 2011).

Impact on Disease Severity and Treatment Adherence

One might assume that as the severity of a child or loved one's disease increases, the caregiver's quality of life would decrease. While this was found in one study (Driscoll et al., 2009), the opposite was found by Besier et al. (2011). Neri et al. (2016) also refuted Driscoll et al.'s (2009) finding, reporting that caregiver burden was independent of the child's clinical findings. On the other hand, a study by Wong and Heriot (2008) found that caregiver despair increased as disease severity in the individual with CF increased.

Smith et al. (2010) found that mothers with increased levels of depression and subsequent decreases in quality of life had children who were less adherent to treatments than children with non-depressed mothers. For example, Smith et al. reported poor

adherence of airway clearance in 41% of the population studied. The same study also measured parent-child relationship security by use of a 17-item relatedness questionnaire and found that children who had a secure relationship with their fathers were 4.7 times as likely to have good adherence with airway clearance ($p < .05$). A study by Barker and Quittner (2016) analyzed parental depressive symptoms with pancreatic enzyme compliance and weight outcomes in children with CF ($n = 83$) aged 1 to 13 years. Fathers represented 12% of the parental sample studied. Although depressive symptoms were found at rates similar to those in other studies (30%), the main finding from this study was that depressed parents were less adherent than non-depressed parents with their children's enzyme therapy (34.8% versus 48.5%). Children who had adherence > 50% had an average gain in weight z-score across 3 months of 0.5 versus -0.1 for children who were less adherent (< 33%). This is an important factor in CF because proper nutrition is closely related to maintaining adequate lung function, which is ultimately related to overall survival.

Financial Stress

Families of patients with CF are also under considerable financial stress. In their cost analysis study, Van Gool, Norman, Dlatycki, Hall, and Massie (2013) found the annual cost of CF ranged from $10,000 to $33,700, depending on disease severity. Another cost analysis of CF in Europe and the United States reported lifetime costs of CF to be estimated at $1.9 million per person (Angelis, Tordrup, & Kanavos, 2015). A study by Briesacher, Quittner, Fouayzi, Zhang, and Swensen (2011) documented a 61%

increase in the costs of private insurance and subsequent care of patients with CF from 2001 to 2007. Studies in this integrated review also mentioned financial stress as a factor impacting the quality of life of caregivers and families (Besier et al., 2011; Driscoll et al., 2009). The CFF (2016) reported that 45% of patients with CF require coverage by Medicaid due to numerous and high-cost medications. Patients are living longer and more medications are being discovered to treat the disease, and as a result, care now costs more. Many families are under significant economic burden, and there are many financial implications they face daily to keep their chronically ill family member as healthy as possible.

Implications for Nursing Practice

Several elements clearly warrant additional attention in this patient population. First, proper studies of mental health screening need to be performed. This action will be a major undertaking for many CF centers that do not have a psychologist as a part of the CF team and will likely rely on nurses to screen. Second, proper referrals need to be assessed to enable affected patients and families to obtain appropriate attention for mental health issues. Third, more studies that evaluate the entire family unit and focus on male caregivers, such as fathers, would advance the scientific body of knowledge. Fathers were poorly represented in all of the studies reviewed, primarily because they are rarely the primary caregiver. Multiple studies have demonstrated that fathers who are assessed are at or near the same risk levels of mothers who are evaluated, yet they receive significantly less attention. It makes sense that a father's quality of life is correlated to the

mother's quality of life, both of which are correlated to the ill child and perhaps other family members. This integrated review of the literature shows that chronic issues impact the entire family and not just the child (or person) with CF.

Conclusion

Quality of life for the caregiver of a patient with CF is a multifactorial issue that also affects the patient and the rest of the family. Quality of life centers on one's general satisfaction with life and expectations for daily living (CDC, 2016). The adverse effects of CF on a caregiver's quality of life are well documented. A correlation exists between anxiety, depression, and poor quality of life, and the rates of these mental health issues are higher in the CF caregiver population than in the general population. A correlation also exists between male and female depression and anxiety among partners taking care of the same child. There is also a link between maternal anxiety and the patient's anxiety. Maternal depression and the security of the father-child relationship have been attributed to the child's adherence to treatment regimens. Other factors that affect quality of life include coping mechanisms and financial stress on families. Few CF centers routinely screen for mental health issues and overall quality of life, and few caregivers or patients seek mental health referrals or resources. The new recommendation from the CFF includes annual screening of both patients and their caregivers for depression and anxiety (Quittner et al., 2016).

In sum, medical care of patients with CF has improved profoundly over the past 20 years, from a focus on symptoms and preparation for an early death to a focus on

helping patients enjoy longer, fuller lives. As a result, the quality of life experienced by parents and others engaged in a complex and ongoing regimen of care has become a significant issue. While much has been learned, much remains to be discovered about the best way to treat and care for patients with CF and their families. Nurses are at the center of all aspects of care for patients with CF. Nurses are afforded a perfect opportunity to advocate for both the families and the patients to ensure that psychosocial screening takes place on a yearly basis, with referrals and follow-up as appropriate.

Declaration of Conflicting Interest

The authors declare that there are no conflicts of interest.

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Table 2.1. Articles included in the integrated literature review

Author	Sample/Setting	Outcome/Findings
Barker & Quittner (2016)	83 parental CGs from 3 CF centers in Florida	Prevalence of depression in CGs using CES-D. Further attempts to correlate depression to adherence to CF patient therapy, PERT, & correlate adherence to weight gain.
Besier & Goldbeck (2011)	162 adolescents with CF and parental CGs in 22 German CF centers	Prevalence of anxiety and depression/healthcare utilization were measured using HADS and CES-D.
Besier et al. (2011)	650 parents of children with CF in 32 German CF centers	Prevalence of anxiety/depression in adolescent patients with CF and CGs was measured using HADS and CES-D.
Driscoll et al. (2009)	122 CGs in a single CF center in the U.S. (Ohio)	Rates of anxiety and depression among CGs were determined. Rates between CGs' anxiety/depression & QOL were assessed. HADS, CES-D, &

		CQOL-CF were used.
Neri et al. (2016)	225 CGs	Depression & vocational participation (presenteeism, absenteeism, sick leave, family leave, reduced work hours) related to CGBSI.
Quittner et al. (2014)	4,102 CGs (F = 3,127; M = 975) in 9 countries (Europe, U.S., & Turkey)	Anxiety & depression in M & F. Anxiety and depression also measured in adolescent and adult patients with CF. Anxiety measured by HADS. Depression by CES-D.
Smith et al. (2010)	39 children, 21 F, 12 M in a single CF center in NY	Depression in patients and CGs was assessed with CDI and CES-D. Anxiety relationship between child or parental depression with AtACT was examined. Status of parent/child relationship and rate of depression was assessed.
Wallenwien et al. (2017)	260 parents, 40 of which were single	QOL in single vs. partnered parents using Ulm QOL inventory

		for parents
Wong & Heriot (2008)	35 CGs of children with CF, ages 5–12 years, in Sydney, Australia, support group	Parental coping, parental vicarious hope and vicarious despair on coping, and parental and child adjustment were investigated. Distinctions between coping, vicarious hope and despair were assessed by VFS.

Note. AtACT = adherence to airway clearance therapy; CDI = Child Depression Inventory; CES-D = Center for Epidemiological Studies Depression Scale; CF = cystic fibrosis; CGBSI = Caregiver Burden Strain Inventory; CGs = caregivers; CQOL-CF = Caregiver Quality of Life for Cystic Fibrosis Scale; F = female; HADS = Hospital Anxiety Depression Scale; M = male; PERT = pancreatic enzyme replacement therapy; QOL = quality of life; R = relationship; SDHS = Short Depression Happiness Scale; VFS = Vicarious Futurity Scale.

Table 2.2. Instruments measuring quality of life of caregivers of patients with CF

Instrument	Description	Study in which instrument was used in this review
Child Depression Inventory (CDI)	CDI screens for childhood depression. CDI has high internal consistency (coefficients = 0.71–0.89), t/rtrc: 0.74–0.83). Cαc: 0.86.	Smith et al. (2010)
Center for Epidemiological Studies Depression Scale (CES-D)	CES-D screens for depression. CES-D has a Cαc of 0.85–0.90.	Barker & Quittner (2016) Besier & Goldbeck (2011) Bessier et al. (2011) Driscoll et al. (2009) Quittner et al. (2014) Smith et al. (2010)
Caregiver Burden Strain Inventory (CGBSI)	CGBSI is a tool used to measure caregiver strain. Correlation with full Depression Happiness Scale (DHS) r = .90. Short Depression Happiness Scale (SDHS) Cα 077–0.92.	Neri et al. (2016)
Caregiver Quality of	Screening tool used in CF to	Driscoll et al. (2009)

Life-CF (CQOL-CF)	measure caregiver QOL. CQOL-CF has Cαc for females = 0.94 and males = 0.84.	
Hospital Anxiety & Depression Scale (HADS)	HADS is widely used screening tool for anxiety and depression. HADS has strong internal consistency, with Cαc of 0.82 for HADS-anxiety and 0.83 for HADS-depression. Sensitivity & specificity 0.80.	Besier & Goldbeck (2011) Besier et al. (2011) Driscoll et al. (2009) Quittner et al. (2014)
Ulm Quality of Life (QOL) Inventory for Parents	Ulm QOL is a QOL tool for parents of chronically ill children. Ulm QOL inventory for parents has a Cα = 0.94.	Wallenwein et al. (2017)
Vicarious Futurity Scale (VFS)	VFS is the hope and well-being scale a parent feels for their child. VFS reliability and validity have been demonstrated using the Cαc for vicarious hope and vicarious despair of 0.83, t/rt of 0.63 and 0.76, respectively	Wong & Heriot (2008)

CHAPTER III

METHODOLOGY

Research Design

A qualitative research design was used in this study. Qualitative studies utilize an emergent design by making decisions about the study as the study progresses by continuously reflecting on the learnings from the study (Polit & Beck, 2012). Phenomenology was the qualitative research tradition that was used in this study. Phenomenology was founded by Edmund Husserl and has roots in both psychology and philosophy (Audi, 2015). The goal of phenomenology is to understand the lived experience of people who experience a particular phenomenon (Polit & Beck, 2012). This study utilized descriptive phenomenology because there is limited information on the experience of fathers who have children with CF, and the aim was to understand their experiences as they are lived, which is the goal of phenomenology (Polit & Beck, 2012).

Setting

Participants for this study were recruited from two CF centers in Texas. Institutional Review Board (IRB) approval was obtained from Texas Woman's University and the participating hospitals and university (see Appendix A and B). Once IRB approval was obtained from the institutions, the researcher worked with employees at the sites to identify fathers who would be coming to clinics with their children. Once identified, the researcher contacted each father to see if he was interested in participating

in the study. If the father was interested, the researcher went over the intent of the study and the inclusion criteria for participation in the study (see Appendix C). Once informed consent was obtained, the semistructured interview was completed, using an interview guide (see Appendix D). The interviews were conducted at locations that were convenient for the participants. This included meeting areas such as at the clinic, at churches, or in the homes of the participants. In cases where it was inconvenient for the participant to have a face-to-face interview, a recorded telephone interview or skype interview was conducted. Polit and Beck (2012) reported that phenomenological studies should be conducted in a setting that is comfortable and private for the participant and one that decreases the risk of loss of confidentiality.

Sampling Strategies

Phenomenological studies are usually small, with 10 participants or fewer who have diverse backgrounds and the phenomenon being studied in common (Polit & Beck, 2012). A phenomenological study in the literature that assessed the lived experience of being a father of a CF patient in Ireland had a target sample size of 10 participants but only enrolled eight participants (Hayes & Savage, 2008). This study limited the age of the child to 1–6 years. Given this information, the target sample of this study was 25 participants, and the target age of the child was newborn to 17 years of age. Recruitment was stopped once data saturation was considered to be achieved. Data saturation is the point at which the researcher feels adequate information has been received and only redundant information is obtained when new participants are added (Polit & Beck, 2012).

Criterion sampling was used in this study. Criterion sampling selects cases that meet a predetermined criterion focused on the phenomenon of interest (Polit & Beck, 2012). In this study, fathers of CF children (children less than 18 years of age) were recruited.

Inclusion criteria for the study included fathers of CF patients who were being treated at a CF center in Texas. The father had to be 18 years of age or older and either the biological or adoptive father. The child needed to have a diagnosis of CF from a CF center and had to be less than 18 years of age. The father had to be able to speak and understand the English language. An exclusion criterion included anyone who was not the father, including stepfathers, grandfathers, and uncles. Also excluded were fathers who did not speak and understand English or who did not have a CF child under the age of 18 or who were under the age of 18 themselves. Participants were reimbursed for parking expenses incurred during the interview and were given a $20 gift card at the completion of the study.

Data Collection

Data were collected in two parts. The first was the collection of demographic data (Appendix E). The second part was data collection congruent with descriptive phenomenology studies and is further described in the next paragraph. The demographic data included age, ethnicity, marital status, highest education level attained, and employment status. Additional demographic information included the total number of children the participant had with CF, if the child with CF was a biological or adopted

child, the gender of the child with CF, the educational level of the child with CF, the current lung function status of the child with CF, and the current age of the child with CF.

The second part of data collection in descriptive phenomenological studies involves a four-step process. The four steps include bracketing, intuiting, analyzing, and describing (Polit & Beck, 2012). The steps of bracketing and intuiting are directly affiliated with data collection. Bracketing occurs when the researcher acknowledges preconceived beliefs that he or she has about the phenomenon being studied; this was done in this study. Although bias can never be fully eliminated, bracketing is done in an attempt to limit bias in the study (Stahl, Taylor, & Hill, 2011). For example, the researcher may have experience with fathers of CF children and think they are less vested in their child's care compared to the mothers. If this were the case, the researcher would need to acknowledge this bias. The second step, intuiting, is when data are collected from the participants. Data were collected in one-on-one semistructured interviews. Polit and Beck (2012) stated that interviews are an important data collection tool in phenomenology studies because they are the very best way for researchers to capture the story of the participants. The semistructured interview questions were open-ended questions attempting to elicit information from participants on their experiences of being fathers of children with CF. One hour was set aside for the interviews, but some took longer. The interviews occurred at a place that was convenient and comfortable for the participants. The researcher worked to build rapport and develop trust with the participants by talking with participants about the study and their child before beginning

the interviews. Polit and Beck reported that data quality will be compromised if participants do not feel they can trust the researcher. These interviews were recorded so the researcher could refer to them later for refreshing and consistency. Because of the length of the interviews and the amount of data being collected, recording the interviews was important to ensure accuracy (Polit & Beck, 2012).

Data Analysis

The final steps in a phenomenological study are analyzing and describing the data (Polit & Beck, 2012). The data must be carefully read and filed in a manner that is accessible for the researcher (Glesne, 2011). The narrative structure of this study involved telling the story of the lived experiences of fathers of CF children. This narrative led to the emergence of major themes from the interviews with the participants. Polit and Beck (2012) described a similarity and contrast principle when conducting interviews and stated that researchers should be alert and looking for similarities and differences immediately and throughout the research process. For example, if the experience of the majority of fathers was guilt associated with a familial genetic disease, that should be noted.

Polit and Beck (2012) described data analysis in phenomenological studies as often fracturing the story down and then rebuilding the story into a holistic narrative. Giorgi described one analytical method that is commonly used in descriptive phenomenology studies (Polit & Beck, 2012). The Giorgi (1985) analytical method was used in this study and involves four distinct steps: (1) thoroughly reading the protocols,

(2) formulating units of meanings from descriptions of the participants in the study, (3) articulating insights from each of the units, and (4) integrating and synthesizing the meaning units into statements that are consistent with the experience of the participants (this is referred to as the "structure of the experience") and can be explained on a specific or a general level.

Due to the level of interaction that must take place between the researcher and the participants in a qualitative phenomenological study, open and honest communication is critical. Another key factor in the success of the study is the ability of the researcher to recognize important cues in body language and communication of the participants. From the interactions, the researcher formed important themes and contrasts from the study participants that led to the conclusions of the study. Once the data were thoroughly analyzed using the Giorgi approach, common themes emerged from the data set. These common themes then described the structure of the lived experiences of CF fathers.

Ethical Considerations

Ethical issues are a concern for all research studies and should be carefully considered. Creswell (2013) proposed that many people believe that ethical issues only arise during data collection; however, ethical issues can be a concern at any point in the study. Participant confidentiality is important and was upheld to the highest standard. Since human participants were involved, IRB approval was obtained prior to the start of the study. All data were de-identified and participants were notified that their

participation or choice not to participate in the study would have no bearing on their children's medical treatment at the CF center.

Rigor

Creswell Rigor Approach

Rigor is directly related to how well the research can be trusted (Glesne, 2011). There are various methods to verify rigor. In this study, Creswell's (2013) criteria for rigor were used. Creswell listed eight characteristics that reinforce rigor and stated that a researcher should use at least two of them in any research study to verify rigor: (a) prolonged engagement, (b) triangulation, (c) peer review, (d) negative case analysis, (e) clarification of research bias, (f) member-checking, (g) rich/thick description, and (h) external audits. This study included the criteria of prolonged engagement; clarification of research bias; member-checking; and rich, thick description. An audit trail was also kept and included a journal and all notes taken during after the interview process.

Description of Rigor

An important aspect of Husserl's (1970) descriptive phenomenology is the use of bracketing. Bracketing is acknowledging the researchers' biases to the study before the study begins and as it progresses (Polit & Beck, 2012). Bracketing was used to meet the rigor criterion of clarification of researcher bias. As previously mentioned, semistructured interviews were conducted with all participants to ensure that adequate data were obtained. Prolonged observation is described as the researcher taking the adequate time that is needed with the participants to obtain the data that are needed (Creswell, 2013).

The semistructured interviews lasted approximately one hour so that the researcher could obtain the desired information from the fathers.

The next rigor criterion that was met during the data collection portion of the study was member-checking. Member-checking is when the researcher goes back to the participants and presents the findings of the interpretations as a way to validate the results (Creswell, 2013). Member-checking can be done once the study is complete or, as in this case, as the data are being collected to verify with each participant at the end of the interview that what they wanted to convey was accurately understood. There is some question as to the plausibility of member-checking because participants may not want to disagree with the researcher or may not remember what was said (Polit & Beck, 2012). It was reasonable to expect honest feedback from this population of daily caregivers for their children with CF. Thick descriptions are described by Creswell (2013) as detailed writings that give readers in-depth knowledge of the subject and should allow for "transferability" of the data. Thick descriptions emerged from the data analysis process of this study into themes.

CHAPTER IV

FINDINGS

A Paper Submitted for Publication in the

Pediatric Nursing Journal

The Lived Experience of Fathers Caring for a Child with Cystic Fibrosis

Abstract

Most studies on the quality of life of caregivers of children with cystic fibrosis (CF) have focused on the mental health of mothers, reflecting a biased underlying assumption that mothers are the primary caregivers. The aim of this study was to explore the experience of fathers caring for a child with CF. Twenty fathers of children with CF were studied via a semistructured interview using Husserl's descriptive phenomenology. Fathers were enrolled from two accredited CF centers in Texas. Six themes emerged from the interviews: fathers reported feeling overwhelmed, feeling isolated, experiencing altered family dynamics, actively seeking resources, experiencing financial strain, and feeling hope. Fathers of children with CF reported distressing experiences in connection with their child's diagnosis of CF and during the course of their child's disease but also reported a strong feeling of hope for the future. Practical implications for nurses include screening for anxiety and depression in fathers at the time of CF diagnosis as well as potentially implementing a peer mentoring program for fathers.

Keywords: quality of life, mental health, financial strain, caregiver, resources, cystic fibrosis

Cystic fibrosis (CF) is the most common life-shortening genetic disease in Whites; affecting 70,000 people worldwide (Cystic Fibrosis Worldwide, 2018). CF is caused by pathogenic mutations of the cystic fibrosis transmembrane conductance regulator (CFTR) gene, which encodes a chloride channel that is mainly expressed in the apical surface of epithelial cells. Altered CFTR expression or function causes abnormal flow of electrolytes and water across epithelial membranes, secretion of mucins that exhibit abnormal rheological properties, and a pro-inflammatory state that leads to dysfunction and tissue destruction of many organ systems (Brennan & Schrijver, 2016). Over the last 60 years, diagnostic and therapeutic advances and a multidisciplinary approach to CF care have improved both the quality of life and the survival of people with CF. Currently, the median survival age of CF patients in the United States is estimated to be 47 years, and although CF is a multisystemic disorder, 90% of patients with CF succumb to lung disease (Cystic Fibrosis Foundation [CFF], 2016).

The burden of CF care is high and requires hours of daily treatments, including chest physiotherapy and multiple inhaled and oral medications due to the systemic nature of the condition. Many of these therapies require active caregiver participation, especially if the patient is a child. The burden of treatment and the experiences associated with having a child with a chronic and progressive course of disease that is often punctuated with acute health issues requiring augmentation of therapy or hospital admissions that

have the potential to alter the caregiver's quality of life and mental health (Barker & Quittner, 2016; Driscoll, Montag-Leifling, Acton, & Modi, 2009; Quittner et al., 2016). Most studies of caregivers' quality of life have focused on the mental health of mothers of children with CF, reflecting a biased underlying assumption that mothers are the primary caregivers of a child with CF and suggesting that fathers are not fully involved or somewhat protected from the burden of caregiving. The purpose of this qualitative phenomenology study was to explore the life experiences of fathers caring for their child with CF.

Methods

Descriptive phenomenology. Husserl's (1970) descriptive phenomenology was utilized in this study. Foundational to Husserl's methodology is the belief that a one-on-one interaction with study participants creates a reflection of reality more explicit than what was earlier understood. Another underpinning of Husserl's philosophy is the belief that each person has the ability to influence his or her own environment, an experience that might or might not be different for each participant (Husserl, 2001). The basic tenets of phenomenology corresponded with the goal of this study, which was to explore and better understand the lived experiences of fathers who have children living with CF.

Selection criteria and data collection. Either biological or adoptive fathers of children with CF were studied, provided they were English-speaking and older than 18 years of age and their children were younger than 18 years of age. Fathers were recruited from the CF clinic of Texas Children's Hospital or Children's Hospital San Antonio

(Both Baylor College of Medicine institutions) when their child had a visit scheduled. The child's diagnosis of CF was based on the CFF (2016) guidelines. Once informed consent was obtained from the study participants, they filled out a questionnaire providing demographic information and relevant data about themselves and their child with CF. The first author then interviewed the participants either in person, via FaceTime voice and video calling (Apple), or by phone. The audiotaped interview followed a semistructured format using the following open-ended questions:

1. Tell me about when your child was diagnosed with CF.
2. Tell me about the day-to-day activities of having a child with CF.
3. Let's talk a little bit about how having a child with CF has changed your relationship with others. Tell how it has (or has not) changed the relationship with your wife? Other children? Child with CF? Friends?
4. How has having a child with CF affected your work?
5. What resources have helped you along this journey with a child with CF? What resources would have helped you more?
6. What advice would you give other fathers of CF children?
7. What else would you like to tell me about your experience of having a child with CF?

Data analysis. Transcripts of the interviews were studied by fracturing and rebuilding each story into a holistic narrative using the data analysis method described by Giorgi (Giorgi, 1985; Polit & Beck, 2012). The Giorgi analytical method used to analyze

the data involved four steps: (1) thoroughly reading the transcripts, (2) formulating units of meaning from descriptions of the participants in the study, (3) articulating insights from each of the units, and (4) integrating and synthesizing the units of meaning into statements consistent with the experience of the participants. The study was approved by the Institutional Review Board of Baylor College of Medicine.

Results

A total of 20 fathers were recruited and completed the study. The demographic characteristics of the fathers and their families are described in detail in Table 4.1. The demographic characteristics of the children of the fathers who participated in the study can be found in Table 4.2.

Several themes emerged from the interviews with the fathers about their experiences of caring for their child with CF. These included being overwhelmed, feeling isolated, experiencing altered family dynamics, actively seeking resources, experiencing financial strain, and feeling hopeful.

Being overwhelmed. The majority of fathers felt overwhelmed at the time of their child's diagnosis and subsequently by the burden of treatment, which typically demanded 3 to 4 hours per day. Although this feeling waned over time as the fathers adjusted to caring for their child, they felt increasingly overwhelmed during acute illnesses and hospitalizations. One father stated: "We have no spontaneity anymore due to all the treatments we have to do. It is just too hard." Many fathers spoke with great emotion when discussing the time of their child's diagnosis. The fathers for whom this

was their second child with CF said they were better prepared, but it was still an emotional time.

Feeling isolated. The fathers reported feeling isolated at diagnosis and during the course of the disease. Parents felt that medical equipment and treatment-related activities limited their mobility. One father noted, "I felt like the boy in the bubble. We feel so isolated with our new diagnosis, treatments, dos and don'ts." Although 50% of fathers reported that they had a good support system, the other half of fathers interviewed revealed they either had no social support or reported a change in their social support after their child's diagnosis.

Experiencing altered family dynamics. Most fathers (60%) reported having a stronger relationship with their wives since their child was diagnosed with CF. Others reported exhaustion and stress due to the time and energy spent on the daily tasks of taking care of their child with CF since their child's diagnosis. One father explained: "We knew this was going to be an issue in our marriage, so we acknowledged it early on."

The study participants were involved in many day-to-day caregiving activities. Each family uniquely divided out care in a way that worked best for them. Interestingly, more fathers dealt with health insurance issues and management of gastrostomy tube feedings for their children. Fathers described their family members as "supportive," although that support was described in various ways. More than one-third of parents received hands-on support from their families and described their support as "phenomenal." In other cases, family members provided emotional support but were

unable to provide hands-on support because they lived a distance away or were unable to understand the nature of CF. One father stated, "Our family wants to somehow box CF up. They want to believe that it is somehow like asthma and not something more serious."

Seeking resources. Seeking resources emerged as a theme during the interviews when the fathers were asked about the diagnosis and what has helped them. Eighty percent of fathers reported that their primary support resource was the CF center. Many fathers mentioned different team members, thus highlighting the importance of the multidisciplinary approach of the CF care centers. Forty-five percent of the fathers also talked about their faith and reliance on God as a major factor of support during the time of diagnosis. One participant stated, "We called them all the time [CF center]. Once we got to the CF center, it made all the difference in my son's life."

One-third of fathers mentioned the CFF and social media websites as being helpful resources during their CF journey. Regarding the CFF website, some viewed the research pipeline on the website, while others were actively involved in local chapters. Social media websites included Facebook pages as well as email lists for parents of children with CF. Twenty percent of fathers relied on general Internet searches as a way to obtain information and resources, while a few fathers warned against this method and said that Internet searches were harmful and sometimes took them to websites with incorrect information.

Encouragement from another mentor family with a child with CF was said to be instrumental by one-quarter of fathers interviewed. Fathers talked about how helpful it was to have contact with or be acquainted with another family living with a child with CF. This was especially true during the time of diagnosis for these fathers, and those fathers who did not have this experience also thought it would have been helpful. One father explained, "This was a phenomenal help to us early in the diagnosis as we were trying to understand everything."

Experiencing financial strain. Financial strain was a common theme. Issues related to finances centered on money, health insurance, and work. The fathers stated that CF is expensive. Even if they had good health insurance, the co-pays and deductibles added up over time to a lot of money. Fifteen percent of the fathers specifically said they relied on pharmaceutical assistance programs to help them offset the costs of medications. Fathers also said that insurance was increasingly difficult to navigate, especially for a parent of a newly diagnosed child. Fathers said it was confusing, and it would have been beneficial to have a case worker to help them.

Fathers also spoke in depth about their work or occupation and how it related to their child's diagnosis of CF. Seventy percent of the fathers felt that their employer was supportive or that they had no work issues related to their children's CF. Fifteen percent of those fathers who reported no issues had not told their employer about their children's diagnosis. Ten percent of the fathers who reported their job as supportive did report difficulties or strain at work if their child was hospitalized. For example, one father

noted, “I have attained a level at my company, and as long as I produce, they are fine with me taking off the time I need.”

Thirty percent of fathers reported currently having work-related difficulty due to the child’s diagnosis of CF. As one father explained, “I frequently have to take Family Medical Leave Act [FMLA] to take care of my son, and I feel threatened for doing so.” In addition, 20% of fathers adapted their careers due to the diagnosis of their child. One father noted, “It has affected the employers who I will consider working for.” Another explained,

> I lost my job during the first year of his life when he was hospitalized for five months. I had no ability to focus or concentrate. The last ten years I have a great job that allows me to work remotely and is supportive. This makes me work even harder for them.

Fathers also mentioned changes to their lifestyles due to their child’s diagnosis of CF. Twenty percent of fathers talked about the child’s mother having to quit her job to care for the child. They also told stories of selling land, property, and businesses so they would qualify for programs and insurances. For instance, one father revealed, “We are considering divorce because we would qualify for Medicaid that way.” Another stated, “Become impoverished—work a minimum wage job. It is the only way to be able to get [the help] your child needs. It is backwards.”

Feeling hope. Hope was the final emerging theme from the interviews with the fathers. The majority of the interviews lasted well over an hour and covered many topics,

but every father ended his interview talking about the hope he had for his child's future and the importance of staying positive. Fathers spoke of the importance of being positive and being engaged with their child. All of the fathers spoke of hope related to the improving therapies in CF and the hope they see reflected in their individual child. As one father stated, "Don't let CF be a reason to not do something."

Fathers talked about the importance of taking each day as it comes and letting their child embrace their childhood despite the diagnosis of CF. Conversely, fathers acknowledged the importance of maintaining a treatment regimen and finding the best doctor possible until a cure is found. One suggested, "Don't treat them differently or like they're fragile." Another stated, "We don't miss medicine—it is our number one role—why we exist."

Discussion

We utilized Husserl's (1970) descriptive phenomenology approach and Giorgi's (1985) analytical method to examine the life experience of fathers caring for a child with CF in the United States. During the semistructured interviews, study participants expressed intense emotions while recounting their experiences. Most of them were active caregivers and were often involved with specific care tasks. All noted that their lives changed dramatically and that they were impacted by their child's diagnosis. They reported being overwhelmed at the time of diagnosis and with time-consuming treatments later on and reported sometimes feeling isolated and stressed out about work and the financial burden of CF. The majority of fathers reported a strong relationship with their

partners but frequently cited exhaustion. These experiences are similar to the previously reported experiences of high satisfaction with family life and little time to relax among caregivers of children with CF (Besier et al., 2011).

Caring for their child with CF was a major concern of participant fathers. They emphasized the help received from their multidisciplinary CF team. Many fathers called out a person by name—either physician, nurse, dietitian, or social worker—who assisted them with navigating their child's care through either specific health issues or from a health resource standpoint.

The majority of fathers reported no issues with their jobs at this time; however, many had struggled in the past, and more than one-third still struggled. Fathers expressed concern about the increasing financial burden of CF, which is in line with data reported by Briesacher, Quittner, Fouayzi, Zhang, and Swensen (2011). They seemed to understand the complexity of both the disease and the insurance system. The majority of study participants had private insurance. Across the United States, 45% of patients with CF require coverage by Medicaid due to numerous and costly medications (CFF, 2016). Finally, study participants felt optimistic, expressed hope about their children's future, and talked about the importance of being engaged with their child. This was the first known qualitative study exclusively conducted on fathers of CF patients in the United States; there was a smaller study conducted on fathers of preschoolers with CF in Ireland (Hayes & Savage, 2008). The experiences reflected in this study seemed to be somewhat the same as in that study reported by Hayes and Savage (2008), in which Irish fathers

emphasized the importance of living day-to-day. Some noted differences between the life experiences of the Irish fathers in that study 10 years ago and this current study of U.S. fathers may be the result, in part, of differences in the age and health status of the children and new treatment options for CF (Hayes & Savage, 2008).

Limitations. This study had some limitations. Data collected in a single interview from study participants whose life experiences might have been different from those who either were not willing to be enrolled or could not participate in the study were a potential source of bias. Moreover, anxiety and depression are highly prevalent in parents of children with CF (Barker & Quittner, 2016; Quittner et al., 2016), but study participants were not screened for these mental health issues, and this was another shortcoming of the study.

Nursing Implications

The data obtained in the present study indicate opportunities for potential interventions for fathers and the entire family at critical times, such as diagnosis and later during the course of CF. Study participants benefited from the interaction with other fathers of children with CF. Parental mentoring programs have been successful in other pediatric chronic illnesses, such as Type 1 diabetes. One such study revealed fathers were the most impacted by a peer mentoring program (Sullivan-Bolyai, Bova, Lee, & Gruppuso, 2011). Fathers reported higher confidence levels managing their children's diabetes than those in the control group when paired with a peer mentor over a period of a year (Sullivan-Bolyai et al., 2011). Fathers also identified the peer mentor as someone

they felt comfortable talking to about disease management and day-to-day activities (Sullivan-Bolyai et al., 2011). That research, coupled with the findings of this study, suggests that it would be worth evaluating such a program for fathers of children with CF.

In conclusion, the findings from this research have practical implications for nurses caring for CF patients today. The CFF and the European Cystic Fibrosis Society have issued guidance suggesting that patients with CF and their parental caregivers be screened for depression and anxiety yearly (Quittner et al., 2016). Their consensus statement lays out an algorithm for screening and treatment and discusses the importance of community referral when appropriate (Quittner et al., 2016). The findings of this study bring to light the importance of fathers caring for their children with CF, as well as the impact that CF has on fathers. While mothers will always play an integral role in the life of children, the role of the father in the treatment of children with CF can no longer be overlooked. The role is relevant because the life experience and mental health of CF caregivers have been shown to influence adherence to CF health maintenance therapy (Barker & Quitter, 2016; Smith, Modi, Quittner, & Wood, 2010). Fathers in this study emphasized the importance of the CF care team as a resource, which creates opportunity for additional studies and interventions. Finally, fathers expressed a strong sense of hope for their children and their children's future and expressed how important that sense of hope was to them as they journey alongside their child with CF.

Funding: This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Acknowledgements: We would like to acknowledge the fathers of patients with CF who were willing to share their stories and experiences of raising their children. We are forever grateful and humbled by these great men and fathers.

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Table 4.1

Demographic Characteristics of the Fathers of Children With Cystic Fibrosis

Demographic	No.	%
Age, y		
18–25	0	0
26–34	7	35
35–44	8	40
45–-54	5	25
> 55	0	0
	M = 38.7	
Ethnicity		
White	16	80
Hispanic	3	15
Asian Indian	1	5
Marital status		
Married	19	95
Single	1	5
Divorced	0	0
Are you married to the mother of child with CF?		
Yes	18	90
No	2	10
If not, are you co-parenting?		
Yes	2	10
Education level		
High school	2	10
Some college	9	45
Bachelor's degree	7	35
Master's degree	1	5
Postdoc/PhD/MD	1	5
Employment status		
Unemployed	1	5
Employed full-time	19	95
Number of children with CF in the family		
1	16	80
2	3	15
3	1	5
Number of children without CF in the family		
0	4	20
1	9	45

2	4	20
3	1	5
4	2	10
Health insurance status		
Private through employer	16	80
Private bought outright	1	5
Medicaid/Medicare	3	15

Note. $N = 20$; CF = cystic fibrosis.

Table 4.2

Demographic Characteristics of the Children With Cystic Fibrosis Whose Fathers Participated in the Study

Demographic	No.	%
Gender of child with CF		
Male	14	70
Female	6	30
Age of child with CF, y		
0–2	5	25
2–5	2	10
6–11	8	40
12–17	5	25
	M = 7.6	
School grade of child with CF		
N/A	7	35
K–5	8	40
6–8	3	15
9–12	1	5
College	1	5
Genotype of child with CF		
DF508/DF508	14	70
Other	6	30
Last known lung function of child with CF (FEV_1)		
Too young to perform	8	40
Unknown	1	5
> 70%	9	45
< 70%	2	10
	* FEV_1 of father-reported lung function: *M* = 102.5% (55-130)	
Number of hospitalizations in past year of child with CF		
0	12	60
1	7	35
2	0	0
3	1	5

Note. *N* = 20; CF = cystic fibrosis; FEV_1 = forced expiratory volume in 1 s.

CHAPTER V

CONCLUSION

Cystic fibrosis is a chronic, complex disease that involves the active participation of a caregiver if the patient is a child. The role of mothers in the care of CF patients has been previously well described. In this study, the experience of fathers caring for their children with CF was examined using Husserl's (1970) descriptive phenomenology. Fathers were mostly recruited in clinics as they came in with their children. There were many key findings from this study.

Demographics

The mean age of the fathers interviewed was 38.7 years and 80% were White, which is consistent with the disease of CF (CFF, 2016). Interestingly, 90% of the fathers interviewed were married to the mothers of the children with CF. This is in contrast to the national divorce rate of 40–50% in the United States (APA, 2018b). Seventy percent of the fathers recruited were fathers of boys, and 30% were fathers of girls. Three of the fathers recruited who were fathers of boys also had older girls (> 18 years of age) with CF. Fathers of children across age groups were recruited.

Education in the study group was representative of all education levels. Specifically, 45% of fathers reported finishing some college, while 35% reported finishing college. The remaining ten percent of fathers reported finishing high school, while 10% reported finishing a master's degree or higher. In terms of employment, 95%

of fathers reported being employed full time. Families of the fathers studied were well insured, with 85% having private insurance and 15% having Medicaid or Medicare. This is in contrast to information from the CFF (2016), which reported 45% of CF families having Medicaid or Medicare.

The children of the fathers studied were well represented across age groups from 0–17 years, with a mean age of 7.6 years. The majority of the children (70%) were homozygous for the deltaf508 mutation, which is the most common CF-causing mutation and is a cause of serious CF disease (CFF, 2016). The mean lung function (as reported by pp FEV_1) was 102.5% of the participants' children. Note that 40% of the children were too young to perform lung function tests. The fathers reported that 60% of the children had not experienced a hospitalization within the last year, while 35% had experienced one and 5% had experienced three.

Findings

Six themes emerged from this study: being overwhelmed, feeling isolated, experiencing altered family dynamics, actively seeking resources, experiencing financial strain, and feeling hope. These themes were discussed at length in Chapter 4.

Future Research

Multiple opportunities exist for future research related to fathers and families in general concerning the care of children with CF. While it is a strength of this study to have enrolled fathers with children across age groups, one area of suggested research would be to follow fathers longitudinally from the time their child is diagnosed with CF

for screening and evaluation of mental health problems, particularly since the fathers interviewed told harrowing stories of their children's diagnoses and their struggles during that period, as discussed in Chapter 4. Another recommendation for further research is a mentoring program for fathers or male caregivers since a few fathers interviewed reported that having another father of a CF patient as a mentor at the time of diagnosis (either formally or informally) was a great asset, as also discussed in Chapter 4.

An area prime for research is a family-centered mental health intervention program in the CF centers. Themes of feeling isolated and being overwhelmed emerged from this study. In addition, descriptions are common in the literature about the impact on parents related to a child's diagnosis and the daily care of CF, as evidenced by increased rates of anxiety and depression (Barker & Quittner, 2016; Quittner et al., 2016). The stage has been set for the next steps in mental health assessment and treatment of CF caregivers.

In conclusion, the findings from this research have practical implications for professionals working with patients with CF. The CFF and the ECFS have issued guidance suggesting that patients with CF and their parental caregivers be screened for depression and anxiety yearly (Quittner et al., 2016). The consensus lays out an algorithm for screening and treatment and discusses the importance of community referral, when appropriate (Quittner et al., 2016). The findings of this study bring to light the importance of fathers caring for their children with CF, as well as the impact that CF has on fathers. Fathers emphasized the importance of the CF care team as a resource, which creates

opportunity for additional studies and interventions. Many advances have been made in caring for CF patients, and research focused on finding the cure continues. As a result, CF patients are living longer than ever, with an average life expectancy of 47 years today (CFF, 2016). This brings a great sense of hope but also financial concerns for families since the cost of CF is rising exponentially and many families need to turn to public assistance to care for their children (Briesacher et al., 2011; CFF, 2016). Finally, fathers expressed a strong sense of hope for their children and their children's future, and healthcare providers must remember how important hope is to this group when carrying out the healthcare of their children.

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APPENDIX A

TWU IRB Approval Letter

Institutional Review Board
Office of Research
6700 Fannin, Houston, TX 77030
713-794-2480
irb-houston@twu.edu
https://www.twu.edu/institutional-review-board-irb/

DATE: May 31, 2018

TO: Ms. Jana Shardonofsky
Nursing

FROM: Ms. Tracy Lindsay, Director of Operations
Office of Research & Sponsored Programs

Re: Institutional Authorization Agreement (IAA) Updated for The Lived Experience of Fathers Caring for their Children with Cystic Fibrosis (Protocol #: 19291)

An IAA for the above referenced study between Texas Woman's University and Baylor College of Medicine was processed as an expedited study. The Baylor College of Medicine IRB is the designated IRB providing the review for this study. According to our records, this protocol was originally approved by the Baylor College of Medicine IRB on 8/22/2016. The TWU IRB has received an updated approval letter and has revised our records to indicate that the most recent approval date is 5/24/2018.

A current protocol file with all correspondence between the researcher and the Baylor College of Medicine IRB must be maintained at TWU. Therefore, you are required to place on file any documentation regarding this study including modifications, extensions, notifications of adverse events, etc.

If you have any questions, please contact the TWU IRB.

cc. Dr. Anita Hufft, Nursing
Dr. Sandra Cesario, Nursing
Graduate School

APPENDIX B

Baylor IRB Approval Letter

May 24, 2018

PETER W HIATT
BAYLOR COLLEGE OF MEDICINE
PEDIATRICS: PULMONARY

H-38891 - THE LIVED EXPERIENCE OF FATHERS CARING FOR THEIR CHILDREN WITH CYSTIC FFBROSIS

APPROVAL VALID FROM 5/24/2018 TO 5/23/2019

Dear Dr. HIATT

The Institutional Review Board for Human Subject Research for Baylor College of Medicine and Affiliated Hospitals (BCM IRB) is pleased to inform you that the research protocol n
Board 1.

The study may not continue after the approval period without additional IRB review and approval for continuation. You will receive an email renewal reminder notice prior to study e
beyond the expiration date.

Please be aware that only IRB-approved informed consent forms may be used when written informed consent is required.

Any changes in study or informed consent procedure must receive review and approval prior to implementation unless the change is necessary for the safety of subjects. In additio
and significant information that may impact a research participants' safety or willingness to continue in your study.

The BCM IRB is organized, operates, and is registered with the United States Office for Human Research Protections according to the regulations codified in the United States Cod

APPENDIX C

Telephone Script

Hello, my name is Jana Shardonofsky, and I am a doctoral student in the nursing program at Texas Woman's University. Thank you for your interest in the research study I am conducting on the experience of fathers caring for their children with cystic fibrosis. Do you have a few minutes to talk about participating in the study?

- If no:
    - I ask "When will you have time to talk?" or ask
    - "Are you still interested in participating in this study?
    - I would then end the call.
- If yes:
    - "I need to collect some personal information about you to see if you qualify for this study. Your taking part in this phone call is completely voluntary. Your information will only be seen by me and my faculty advisor at Texas Woman's University. We try to make sure the information we collect from you is kept private and used only for the research study we are discussing."

If the phone call will continue:

- "I need to ask you a few questions to see if you meet the criteria for the study."
    - Are you the biological or adopted father of a child with cystic fibrosis?
    - Is your child under the age of 18?
    - Are you over the age of 18?

- Are you able to speak and understand the English language?
- Would you be able to meet for a face-to-face interview or conduct an interview on the phone/skype?

- If no:
  - "I appreciate your interest in the study but you do not meet the criteria for the study. Thank you for taking the time out of your day to call me. Good bye."
- If yes:
  - "It seems that you do meet the criteria for the study. Can I take a few more minutes of your time to tell you what this study would entail?"
- If yes:
  - "Again the purpose of this study is to understand the experience of the father caring for a child with cystic fibrosis. This research study would require that you provide some general information about yourself which I would keep private in a locked file cabinet in my office. The main requirement for this study is an approximate one hour interview with me to tell me about your experience caring for your daughter or son with cystic fibrosis. I will take notes during the interview and will audio-record the interview so I can go back and make sure I have the most accurate account of what you told me. We would meet one time and there may be one follow-up call."

- "One potential risks in this study is loss of confidentiality. Confidentiality will be protected to the extent that is allowed by the law. The interview will be held in a private mutually agreed upon location. A code name, not your real name will be used during the interview. No one but my faculty advisor and I will know your real name. The files from your interview will be kept in a locked file in my office. There is a potential risk of loss of confidentiality in all email and internet transactions.
- Another potential risks in this study is loss of time. Including the time you are talking to me today, this study may take up to 2 hours of your time. One hour of that will be the interview, 15 minutes today, 15 minutes filling out paper work and the potential for a 30 minute follow-up phone at the end of my study to check the accuracy of my findings with you.
- Another potential risk in this study is emotional discomfort. I will do everything to make you comfortable but some topics may make you feel emotional distress. If you become tired or upset at any time we can take breaks. You can also stop answering questions and stop the interview at any time. If you feel you need to talk to a professional about your emotional distress, I will provide a list of resources."

- **“**Benefits of the study include a $20 gift card at the completion of the interview as well as being reimbursed for any parking expenses incurred during the interview. There are no other benefits are costs to you.”

At the completion of the explanation:

- “Do you have any additional questions about the study you would like to ask?”
- “Do you think that you are interested in participating in this pilot study?”
- “Thank you for your time today.”

APPENDIX D

Interview Guide

1. Tell me about when your child was diagnosed with CF.
2. Tell me about the day-to-day activities of have a child with CF.
3. Let's talk a little about how having a child with CF has changed your relationships with others.
    a. Tell me how it has changed your relationship with your: wife?
    b. Other children?
    c. Child with CF?
    d. Friends?
4. How has having a child with CF affected your work?
5. What resources have helped you along this journey with a child with CF? What resources would have helped you more?
6. What advice would you give other fathers of children with CF?
7. What else would you like to tell me about your experience having a child with CF?

APPENDIX E

Demographic Intake Form

Age:_____

1. Ethnicity/Race:

( ) White/Caucasian

( ) African American/Black

( ) Hispanic (Mexican/Mexican American, Puerto Rican, Cuban or another Latino or Spanish Origin)

() American Indian or Alaska Native

( ) Asian Indian

( ) Japanese

() Native Hawaiian

() Chinese

() Korean

() Guamanian or Chamorro

() Filipino

() Vietnamese

() Samoan

() Other Asian

() Other Pacific Islander

() Some Other Race

2. Marital Status: ( ) Married

   ( ) Single

   ( ) Divorced

   ( ) Widowed

3. If married, are you married to the mother of your child with cystic fibrosis?

   _____

4. If you are not married to the mother of your child with cystic fibrosis, are you co-parenting your child with CF? _____

5. Education Level:

   ( ) Did not graduate high school

   ( ) High school

   ( ) Some college

   ( ) Associates degree

   ( ) Bachelor's degree

   ( ) Master's degree

   ( ) Doctorate (PhD, MD or similar)

6. Employment Status:

   ( ) Unemployed

   ( ) Student

   ( ) Employed Part-Time

   ( ) Employed Full-Time

( ) Retired

7. Number of Children:_____

8. Number of children with cystic fibrosis:_____

9. How long ago was your child diagnosed with CF? _____

10. Is your child with cystic fibrosis a biological or adopted child? _____

11. Gender of your child or children with cystic fibrosis:_____

12. Age of your child or children with cystic fibrosis:_____

13. If child with CF is in school, what grade?_____

14. Genotype of child with CF:_____

15. Number of hospitalization child with CF had in the past year:_____

16. Last known lung function of child with CF (if performed):_____

17. Age and Gender of Children in the Family without

    CF_____

18. Insurance status: (Check Primary Insurance)

    ( ) Private insurance through employer

    ( ) Private insurance bought outright

    ( ) Tricare/Military Insurance

    ( ) Medicaid/Medicare or

    ( ) Not insured

19. Check Secondary Insurance (If Applicable)

    ( ) Private insurance through employer

( ) Private insurance bought outright

( ) Tricare/Military Insurance

( ) Medicaid/Medicare

APPENDIX F

Manuscript Submission and Publication Release Letters

TWU

Jana Shardonofsky <jshardonofsky@twu.edu>

Quality of Life in CF Care Givers: An Integrated Literature Review

Jackie Massaro <jackie.massaro@ajj.com> Tue, Oct 9, 2018 at 11:45 AM
To: Jana Shardonofsky <jshardonofsky@twu.edu>

Hello Jana,

Received, thank you for sending. Your manuscript is now out for review.

Please don't hesitate to contact me with further questions or concerns.

Kind Regards,

Jackie

From: "Jana Shardonofsky" <jshardonofsky@twu.edu>
To: "Jackie Massaro" <jackie.massaro@ajj.com>
Sent: Saturday, October 6, 2018 7:02:16 PM
Subject: Quality of Life in CF Care Givers: An Integrated Literature Review
[Quoted text hidden]

TWU

Jana Shardonofsky <jshardonofsky@twu.edu>

FW: Jana Shardonofsky Submission

5 messages

Zerangue, Amanda <AZerangue@twu.edu> Mon, Oct 1, 2018 at 9:21 AM
To: "Shardonofsky, Jana" <JShardonofsky@twu.edu>
Cc: "Cesario, Sandra" <SCesario@twu.edu>, "Chowritmootoo, Michan" <MChowritmootoo@twu.edu>

Good Morning, Jana:

Wonderful news! Jackie Massaro, Pediatric Nursing's Editorial Coordinator, has provided permission to use your manuscript as a chapter in your dissertation.

Please see her written permission below.

Please let me know if I may help in any way.

Best,

Amanda

AMANDA R. ZERANGUE, M.L.S | J.D

Manager of Digital Services & Scholarly Communication Librarian

TWU Libraries

P: 940 898 3747 | azerangue@twu.edu

LIBRARIES
TEXAS WOMAN'S UNIVERSITY

From: Zerangue, Amanda
Sent: Monday, October 01, 2018 9:17 AM
To: 'Jackie Massaro' <jackie.massaro@ajj.com>
Subject: RE: Jana Shardonofsky Submission

This is wonderful news! Thank you so much, Ms. Massaro.

Best,

Amanda

AMANDA R. ZERANGUE, M.L.S | J.D

Manager of Digital Services & Scholarly Communication Librarian

TWU Libraries

P: 940 898 3747 | azerangue@twu.edu

From: Jackie Massaro [mailto:jackie.massaro@ajj.com]
Sent: Monday, October 01, 2018 9:15 AM
To: Zerangue, Amanda <AZerangue@twu.edu>
Cc: Jackie Massaro <jackie.massaro@ajj.com>
Subject: Re: Jana Shardonofsky Submission

Dear Amanda,

Thank you for your message.

Ms. Shardonofsky still needs to send her Transfer of Copyright document to the office (can send to my email). However, because she is the author, we will allow Ms. Shardonofsky to include her article as part of her dissertation at no cost and with no complications.

Thank you.

Kind Regards,

Jackie Massaro
Editorial Coordinator
Pediatric Nursing
(856) 256-2300 EX 2344
jackie.massaro@ajj.com

Facebook: www.facebook.com/PediatricNursing

Twitter: www.twitter.com/PedNursing

From: "Zerangue, Amanda" <AZerangue@twu.edu>
To: "Jackie Massaro" <jackie.massaro@ajj.com>
Sent: Friday, September 28, 2018 12:46:12 PM
Subject: Jana Shardonofsky Submission

Good Morning, Ms. Massaro:

I am hoping to help one of the graduate students at Texas Woman's University (TWU), Jana Shardonofsky, with a few copyright questions. She has submitted a manuscript to Pediatric Nursing, and it is under review. She is also using this manuscript as a chapter in her dissertation.

If her article is accepted by Pediatric Nursing, may Ms. Shardonofsky use her original manuscript as a chapter in her dissertation? At TWU, the dissertations may be submitted to Proquest, and must be submitted to our institutional repository (after any applicable embargoes).

Can you help us understand whether Ms. Shardonofsky may retain the right to include her manuscript as a chapter in her dissertation? I reviewed the Author Packet provided, but did not see any information about dissertation reuse or embargo periods.

I appreciate any information you may have.

Best,

Amanda

AMANDA R. ZERANGUE, M.L.S | J.D

Manager of Digital Services & Scholarly Communication Librarian

TWU Libraries

P: 940 898 3747 | azerangue@twu.edu

Chowritmootoo, Michan <MChowritmootoo@twu.edu> Mon, Oct 1, 2018 at 9:28 AM
To: "Zerangue, Amanda" <AZerangue@twu.edu>, "Shardonofsky, Jana" <JShardonofsky@twu.edu>
Cc: "Cesario, Sandra" <SCesario@twu.edu>

Jana

From: "Zerangue, Amanda" <AZerangue@twu.edu>
To: "Jackie Massaro" <jackie.massaro@ajj.com>
Sent: Friday, September 28, 2018 12:46:12 PM
Subject: Jana Shardonofsky Submission

Good Morning, Ms. Massaro:

I am hoping to help one of the graduate students at Texas Woman's University (TWU), Jana Shardonofsky, with a few copyright questions. She has submitted a manuscript to Pediatric Nursing, and it is under review. She is also using this manuscript as a chapter in her dissertation.

If her article is accepted by Pediatric Nursing, may Ms. Shardonofsky use her original manuscript as a chapter in her dissertation? At TWU, the dissertations may be submitted to Proquest, and must be submitted to our institutional repository (after any applicable embargoes).

Can you help us understand whether Ms. Shardonofsky may retain the right to include her manuscript as a chapter in her dissertation? I reviewed the Author Packet provided, but did not see any information about dissertation reuse or embargo periods.

I appreciate any information you may have.

Best,

Amanda

AMANDA R. ZERANGUE, M.L.S | J.D

Manager of Digital Services & Scholarly Communication Librarian

TWU Libraries

P: 940 898 3747 | azerangue@twu.edu

LIBRARIES
TEXAS WOMAN'S UNIVERSITY

Chowritmootoo, Michan <MChowritmootoo@twu.edu> Mon, Oct 1, 2018 at 9:28 AM
To: "Zerangue, Amanda" <AZerangue@twu.edu>, "Shardonofsky, Jana" <JShardonofsky@twu.edu>
Cc: "Cesario, Sandra" <SCesario@twu.edu>

Jana

TWU

Jana Shardonofsky <jshardonofsky@twu.edu>

Manuscript for Submission

Jackie Massaro <jackie.massaro@ajj.com> Mon, Oct 29, 2018 at 10:47 AM
To: Jana Shardonofsky <jshardonofsky@twu.edu>
Cc: Jackie Massaro <jackie.massaro@ajj.com>

Dear Ms. Shardonofsky:

Thank you for submitting your manuscript, "The Lived Experience of Fathers Caring for their Child with Cystic Fibrosis," to be considered for publication in **Pediatric Nursing**. Your manuscript has been forwarded to the editor for review assignment by our manuscript reviewers. Please expect a response in approximately 3-5 months.

In order to process your manuscript, each author of the manuscript must complete and return the attached author packet to me via email. **(A CV for each author will also be required.)** Whenever possible, the corresponding author should send all of this information in one or as few emails as possible. If you have previously submitted this material, you may disregard this request.

If you intend to use any photographs in your article, please use your institution's regular form or request a permission form from Jannetti Publications. The publisher assumes the author has acquired written permission for all photographs and other non-original material to be used in your manuscript, and the author should supply the publisher with copies of those permissions.

Please note that upon your submission to **Pediatric Nursing**, you are stipulating your submission is an original work, and is not currently under consideration by another publication. If you have concurrently submitted this work to another publication, please let me know as soon as possible.

If you should decide to withdraw your manuscript, please notify me. Withdrawn or rejected manuscripts will be destroyed and purged from our electronic system to protect your private information.

Again, thank you for submitting your manuscript to **Pediatric Nursing**.

Sincerely,

Jackie Massaro
Editorial Coordinator
Pediatric Nursing
(856) 256-2300 EX 2344
jackie.massaro@ajj.com
Facebook: www.facebook.com/PediatricNursing
Twitter: www.twitter.com/PedNursing

From: "Jana Shardonofsky" <jshardonofsky@twu.edu>
To: "Jackie Massaro" <jackie.massaro@ajj.com>
Sent: Thursday, October 18, 2018 9:53:01 PM
Subject: Manuscript for Submission
[Quoted text hidden]

APPENDIX G

Signature Page

TEXAS WOMAN'S UNIVERSITY
DENTON, TEXAS

November 6, 2018

To the Dean of the Graduate School:

I am submitting herewith a dissertation written by Jana Shardonofsky entitled "The Lived Experience of Fathers Caring for Their Child With Cystic Fibrosis." I have examined this thesis/dissertation for form and content and recommend that it be accepted in partial fulfillment of the requirements for the degree of Doctor of Philosophy with a major in Nursing Science.

Sandra Cesario
Sandra Cesario, PhD, Major Professor

We have read this thesis/dissertation and recommend its acceptance:

Nina Fredland
Nina Fredland, PhD

Peggy Landrum
Peggy Landrum, PhD

Ainslie Nibert
Ainslie Nibert, PhD, Associate Dean

Accepted:

Carolyn Kapinus, PhD

CURRICULUM VITAE

Jana Shardonofsky
139 Montclair
San Antonio, Texas, 78209
Cell (214) 454-1569
Home (210) 384-4523

I. GENERAL BIOGRAPHICAL INFORMATION

A. Education

Graduate:

May 1996–May 1999	Master of Science in Advanced Practice Nursing Family Nurse Practitioner Program Texas A&M University Corpus Christi, Texas

Undergraduate:

January 1993–August 1994	Bachelor of Science in Nursing School of Nursing University of Texas Health Science Center at Houston Houston, Texas

B. Academic Appointments

July 1999–January 2004	Instructor of Pediatrics, Pediatric Pulmonology Baylor College of Medicine Houston, Texas

C. Experience/Advanced Training

October 2018–Present	Regional Director of Medical Affairs Heron Therapeutics North America-West

August 2015–October 2018	Vertex Pharmaceuticals Associate Director, West Medical Science Liaisons North American Medical Affairs
August 2011–August 2015	Vertex Pharmaceuticals Senior Medical Science Liaison South/Central Region Cystic Fibrosis Field Medical Team
March 2010–August 2011& September 2006–April 2008	Gilead Sciences Director/Associate Director, Medical Affairs Cardiopulmonary Division Western Region and Texas Territory
April 2008–March 2010	Eurand Medical Science Liaison Team Manager Associate Director, Medical Affairs Dallas/Houston, Texas
December 2004–August 2006	Chiron Corporation Medical Science Liaison Pulmonary/Cystic Fibrosis Central and South Regions Dallas, Texas
January–November 2004	Chiron Corporation Area Business Manager Biopharmaceuticals/Pulmonary/Infectious Disease Dallas, Texas
May 1999–January 2004	Pediatric Pulmonary Family Nurse Practitioner Baylor College of Medicine Houston, Texas
January 1998–May 1999	Pediatric Pulmonary Clinical Nurse Baylor College of Medicine Houston, Texas
July 1996–December 1997	Pediatric Pulmonary Nurse Clinician Cystic Fibrosis Nurse (Adult and Pediatric) Scott & White Clinic Temple, Texas
September 1995–July 1996	Staff Nurse/Charge Nurse, Pediatric Unit Scott & White Clinic Temple, Texas

September 1994–August 1995	Staff Nurse/Charge Nurse, Medical Surgical, ICU, ER Coryell Memorial Hospital Gatesville, Texas
February 1994–August 1994	Student Nurse Associate, OB-Float St. Luke's Episcopal Hospital Houston, Texas

D. Certifications/Licenses

Texas State Board of Nurse Examiners, #610404

E. Honors/Awards

Suma Cum Laude Graduate—Master of Science in Nursing, Texas A&M University, Corpus Christi
Magna Cum Laude Graduate—Bachelor of Science in Nursing, University of Texas Health Science Center, Houston
Sigma Theta Tau, International Honor Society for Nurses, University of Texas—Houston Chapter and Texas Woman's University—Denton

F. Other

Nursing Speakers Advisory Panel, Chiron Corporation, 2001
Managed Care Speakers Panel, Prime INC/Chiron Corporation, 2003

II. RESEARCH INFORMATION

A. Research Experience

Research Coordinator, Cystic Fibrosis Epidemiology Study
Scott & White Clinic
Temple, Texas

An International, Multicenter, Uncontrolled, Open Evaluation of Chronic UT-15 Plus Conventional Therapy in Patients with Pulmonary Hypertension: A Continuation Study. United Therapeutics Corporation. Co-Principal Investigator.

B. Publications/Presentations

Alternate Administration Options for Pancreatic Enzyme Products (PEPs): Food on Which PEPs Can Be Administered. Abstract Presentation: American Dietetic Association (ADA). Denver, Colorado. October 2009.

New Directions in the Management of Cystic Fibrosis Disease. National Speaker's Venue for PRIME, INC, March 2003 to 2004.

Medical and Nutritional Management of Cystic Fibrosis Related Diabetes. Sixteenth Annual North American Cystic Fibrosis Conference, New Orleans, Louisiana. October 2002.

Cystic Fibrosis in the Pediatric Patient. Cook Children's Hospital Nursing Continuing Education Conference, Fort Worth, Texas. September 2001.

A Guide for Education and Treatment of Asthma in the Community. Presented to Houston Area Nurses. November 2001, March 2001, November 2002.

The ABC's of Pulmonary Assessment in the Pediatric Patient. The Children's Hospital Association of Texas Conference (CHAT), Houston, Texas. October 2000.

Pulmonary Hypertension. Pulmonary Hypertension Association Fundraiser, Houston, Texas. March 2000.

Flolan and Pulmonary Hypertension Update. Texas Children's Hospital, Houston, Texas. January 2000.

RSV, Croup, and Bronchiolitis. Pediatric Symposium, Scott and White Hospital, Temple, Texas. April 1998.

The Uninsured Population in Central Texas—An Overview, Nursing Research Conference, Scott & White Hospital/Clinic, Temple, Texas. Fall 1997.

Cystic Fibrosis, Tuberculosis, and Bronchopulmonary Dysplasia—An Update, Pediatric Challenges Conference, Scott & White Children's Health Center, Temple, Texas, 1997.

Clark, J. RSV: Its effects on infants and young children. *Nurse To Nurse*, Scott and White Publications, Spring 1997.

Pediatric Grand Rounds, Update on Cystic Fibrosis Conference, December 1996.