AN EXPLORATION OF THE EXPERIENCES OF CAREGIVERS RAISING CHILDREN WITH A CYSTIC FIBROSIS DIAGNOSIS AND THE RELATIONSHIP BETWEEN ANXIETY, DEPRESSION, AND QUALITY OF LIFE: AN ONLINE EXPLORATORY MIXED-METHODS STUDY

A DISSERTATION

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COLLEGE OF PROFESSIONAL EDUCATION

BY

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DEDICATION

This work is dedicated to the young girls out there growing up:

Who are poor and facing food and housing security. Who may have experienced significant abuse and trauma. Who are seen as unpopular. Who have been called “weird,” “different,” or “other.” Who have been told they are “too much” of anything. Who are worried they are not enough. Who wonder what they will make of themselves in this world. Who yearn for normalcy and to fit in. Who are afraid to speak their truth and to be authentically themselves. Who are the “strong” ones in the family used to taking care of others and putting themselves and their needs last.

You will get through this. You are beautiful. You are enough.

Fitting in is BS; weird is wonderful.

Be bold and brave and authentically you. The world doesn’t more need carbon copies of average; it needs exactly who you were meant and created to be. Those who are meant for you will find you; and those who are not will fade away. Choose courage over comfort every day and you will find your way.

If you are that girl now or have been that girl: this is for you. I see you. I too was that girl.

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An extremely special thank you to the participants in this study who contributed their time, knowledge, and personal experiences. My hope is that while we await a cure for this disease, this research will provide a foundation to bring more awareness to CF within the mental health world. You all deserve quality health care where you are not bogged down in explaining the physical aspects and time burden associated with CF over and over and over again. We know CF affects the entire family and your involvement in this study helped to illuminate just how deeply embedded the uninvited guest of CF is within your homes. You were raw, vulnerable, emotional, and real and you have my sincerest gratitude in sharing your experiences. May the 65 roses no longer take our loved one's breath away.

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To my hunky husband, Nick: “You’re my candle in a darkened house. You say, “Relax, I’ll lead the way” – A&B. Thank you for your constant support, encouragement

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have gotten through so much pain and heartache together. I am so grateful we are finally in a good place. Dad, thanks for always being a stubborn rock that I can rely on for sound advice and comfort. Grams & Grampy: you instilled a love of reading, learning, continued curiosity, and travel in me from a very young age, and I am forever grateful. Grams – thanks for the mantra that "only boring people get bored," as it has shaped my path. Grandpa, I miss you so very much, especially and our late-night driveway talks eating Oreo cake and all of our foodie adventures.

To my brothers: thank you for all the love, support, laughter, and memories. Dakota, my sweet middle brother… you left us far too early, little one. The sudden and unexpected loss of your vibrant smile, kindness, thoughtfulness, and warm and comforting hugs is beyond overwhelming. We had the most in common and shared a love of music, dancing, and being at the beach together. No one could ever fill the special place you have always held in my heart. I hope by the time we meet again the stigma surrounding mental health will have diminished greatly and resources for those struggling will be readily available, affordable, and plentiful. I will continue to fiercely advocate for mental health awareness, support, education, and normalizing this process for the rest of my life. May you be at peace and be free to be yourself – until we can laugh and dance together again, I love you so, so, so very much. To my family who are active participants in my life: thank you for enriching my life and existence. I hope the feeling is mutual.

To my best friend, Becca, the strongest mama I have ever met and the inspiration for this research. Thank you for the direction, education, and constant support. Thank you for also piloting this study. Thank you for the virtual long-distance retail shopping dates.

I hope this work leads to positive changes within the community and is a work in which you are proud. To my best girl, Amaya: you are our CF warrior who is not only beautiful on the outside, but the inside as well. Thank you for the laughter and constantly making your mother and I question if our wardrobe and life choices are fashionable. I assure you; they are not!

To my girlfriends (you know who you are!), the best group of women to have in this process - thank you for everything. Since I am the last one to finish because I do things on Hawaiian time (I blame my mother for this LOL), we can now have our massive bonfires and girl's trips to celebrate. Your encouragement over the past few years has been a blessing and I am so grateful to have you all in my life.

If I have forgotten anyone specifically, I apologize.

ABSTRACT

KRISTINA DINGUS KEUHLEN

AN EXPLORATION OF THE EXPERIENCES OF CAREGIVERS RAISING CHILDREN WITH A CYSTIC FIBROSIS DIAGNOSIS AND THE RELATIONSHIP BETWEEN ANXIETY, DEPRESSION, AND QUALITY OF LIFE: AN ONLINE EXPLORATORY MIXED-METHODS STUDY

MAY 2021

This online mixed-methods study was designed to explore, analyze, expand upon the lived experiences of caregivers raising children with cystic fibrosis (CF) and the connection to the systemic influences contributing to their elevated rates of psychological symptoms. A main purpose of this study was to expand specifically upon the TIDES data (Quittner et al., 2014), which found that caregivers in this demographic experience two to three times elevated rates of anxiety and depression than community samples. The hope of this study was to investigate and obtain an understanding of the obstacles this group faces daily while analyzing the data, to give a voice and meaning to participant's reality.

A total of 155 participants completed the survey, predominantly mothers (n = 142) with a good representation from fathers (n = 13). To assess psychological functioning, participants (n = 100) completed four quantitative surveys: Patient Health Questionnaire-9 (PHQ-9), General Anxiety Disorder-7 (GAD-7), Caregiver Quality of Life-Cystic Fibrosis (CQOLCF), and Cystic Fibrosis Questionnaire-Revised (CFQ-R). MANOVA testing found

parents raising adolescent daughters ages 14 and older reported the lowest rates of anxiety and depression and highest quality of life as compared to parents of sons and all other groups. Caregivers raising children from birth to 6 years old experienced elevated levels of anxiety and depression. Mothers reported higher levels of anxiety and depression and lower quality of life, as a whole, while fathers reported greater struggles with symptoms of anxiety than depression.

The present study expanded the quantitative analysis from TIDES by incorporating a qualitative exploration into the lives of caregivers raising a child with CF, with a range of participant responses (56 to 82). Multiple levels of coding found five themes consisting of The CF priority, Resources & Support, Pandemic Amplification of Distress, Natural Skepticism and Cautious Optimism, and Barriers to Seeking and Receiving Mental Health Services. These themes helped to highlight the obstacles caregivers face when considering mental health treatment interventions, specifically stigma and access to therapists. It is recommended that a shift in the current mental health program be assessed to incorporate enhanced and more frequent psychological check-ups for children with CF as well as their caregivers.

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CHAPTER I

INTRODUCTION

Cystic fibrosis (CF) is the most common genetic, life-threatening disease affecting Caucasians worldwide, with approximately 30,000 cases in the United States (Cystic Fibrosis Worldwide, 2019). The following chapter will provide detailed information about cystic fibrosis, its prevalence and impact on individual and relational functioning from a physical, mental, and emotional viewpoint. A brief review of literature will discuss historical and recent data pertaining to the cystic fibrosis population and the impact this life-shortening disease has on rates of anxiety, depression, and quality of life for caregivers raising a child with CF. This chapter will also identify the problem and purpose statements, rationale and significance of the study, theory guiding the research, research questions, tables of definitions, assumptions of the study, delimitations of participants, and a summary of the chapter before moving into the literature review.

Prevalence of Cystic Fibrosis

According to the Cystic Fibrosis Foundation (CFF), CF is the number one progressive, genetic, life-shortening, chronic illness of Caucasians which primarily affects the lungs and digestive system of the individual (CFF, 2014b). Cystic Fibrosis Worldwide (CFWW; 2019) identified approximately 30,000 individuals diagnosed in the United States with approximately 1000 new cases diagnosed annually. A majority (75%) of CF diagnoses are received by the age of two; at this time, 50% of individuals with CF are over the age of 18. The life expectancy of individuals with CF is approximately 37

years if they were born in the year 2000 or later. In 1985 the average life expectancy was 27; in 2009, it was almost 36 (CFF, 2018). Recent data from the 2019 CFF Patient Registry indicates that for individuals born between 2015 and 2019, it is predicted half will live to 46 or older (CFF, 2021a).

CF treatments range daily from nebulizers, vest treatments that encourage mucous production and release, and an assortment of medications and enzymes to help children absorb much needed-nutrients, breathe effectively, and function physically in the most normal way possible (CFF, 2014b). Daily treatments, scheduled doctor appointments and CF care clinic visits, planned and emergency hospital visits, and special considerations for social activities, educational opportunities, and familial obligations are all challenges due to the diagnosed child's survival needs taking precedence; all of these factors will take a toll on the caregivers of the child. Mental health clinicians must ask themselves, "What is the impact of up to four hours of daily treatments for an individual with CF, let alone their caregivers and families?" That is what this study aims to address.

Brief Literature Review

Although chronic illness is not a new phenomenon, CF is a relatively new disease that was discovered in 1939 (Quittner, 1998). Until the late 1980s, very little was known about the cause or potential treatment, let alone a cure. CF is different from other chronic illnesses, as diagnosed individuals have a lower life expectancy rate than that of the well population and other chronic illnesses. As more individuals are diagnosed annually, it is important to understand how the illness affects the entire family unit including caregivers, siblings, and extended familial relationships, to best serve this demographic within the

realm of psychological health. Many individuals with CF or caregivers of children with CF acknowledge the lack of time for individual, couple, or family therapy as so much of their daily lives are focused on the survival and comfort of the child (R. Ellithorpe, personal communication, March 2017). Therefore, it is important to understand the special circumstances for these families so we may effectively and efficiently incorporate mental health services into their protocol of treatments.

Anxiety and depression are the two most commonly diagnosed mental illnesses in the United States (National Institute of Mental Health [NIMH], 2019). Annually, over 40 million individuals are diagnosed with anxiety and 17 million diagnosed with depression (NIMH, 2019). Studies suggest caregiver rates of anxiety and depression are elevated when raising a child with CF compared to the well population (Besier et al., 2011; Cruz et al., 2009; Driscoll et al., 2009; Driscoll et al., 2010; Hodgkinson & Lester, 2002; Quittner et al., 2008; Quittner et al., 2014). The International Depression and Anxiety Epidemiological Study (TIDES) surveyed over 6,000 patients and caregivers across nine countries throughout Europe and the United States (Quittner et al., 2014). TIDES identified increased rates of psychological symptoms including anxiety and depression in adults and adolescents with CF as well as parent caregivers with rates two to three times higher than community samples (Smith et al., 2016). Additionally, TIDES data indicated adolescents were twice as likely to exhibit elevated rates of anxiety and depression when one parent reported symptoms (Smith et al., 2016).

Bryon and Webb (2015) stated the TIDES study is lacking in the connection between lived experiences, context, and systemic influences contributing to the elevated

rates of psychological symptoms within the CF community. The TIDES results received a major critique, “more work is required to better understand the nature of the psychological needs of people with CF, and their families as well as the best service provision to ensure needs are met” (Bryon & Webb, 2015, p. 490). It has also been recently documented that “a key goal is how to maintain mental health across the lifespan in CF, including prevention measures, screening, and psychological or medication interventions” (Garcia et al., 2018, p. 159). In order to best serve this population psychologically, more information is vital to connect the meaning of lived experiences with numerical data to bridge the gap where research is lacking after mental health screenings are performed, including evaluations, interventions, and referrals (Garcia et al., 2018).

Quality of Life

It has been documented that individuals with chronic illness exhibit elevated levels of anxiety and depression and may have diminished levels of quality of life, marital satisfaction, and life satisfaction (Bergsten Brucefors et al., 2015; Besier & Goldbeck, 2012; Besier et al., 2011; Cruz et al., 2009; Driscoll et al., 2009; Driscoll et al., 2010; Hodgkinson & Lester, 2002; Quittner et al., 2008; Quittner et al., 2014). Individuals with CF face daily stressors from medications, medical treatments, and infection control that far exceed other chronic illness and disease. The overwhelming daily needs for survival of the individual with CF place a burden of treatment and care that affects their entire biopsychosocial system creating family dynamics that other chronically ill individuals and families, and the healthy population, do not encounter. The

need to identify the special circumstances and challenges these families face and how it effects their psychological health and emotional wellness is urgent.

Problem Statement

There is an increasing body of literature that focuses on better understanding the state of mental health in parents and caregivers raising children with CF (Driscoll et al., 2009; Garcia et al., 2018; Shardonofsky, Cesario, Fredland, Landrum, Hiatt, et al. 2019b). The findings from this relatively small body of research indicate parents and caregivers exhibit elevated rates of anxiety and depression and lower life satisfaction and quality of life. As this body of knowledge has grown, the CFF has recommended an annual psychological screening of anxiety and depression for all individuals with CF, and their caregivers, at routine clinic visits (Abbott et al., 2015; Garcia et al., 2015; Quittner et al., 2014).

With the recent introduction of annual mental health screenings within CF clinics, it is vital to understand the connection between the numerical data representing rates of anxiety and depression, the daily struggles contributing to the increased rates, the barriers to seeking and receiving mental health care, and beginning a dialogue to identify best practices of treatment and psychological intervention within the mental health community. The CF medical community has established a high quality of integrative and collaborative care within their clinic care teams to address physical health concerns and have recently begun implementing annual psychological screenings for anxiety and depression of individuals with a CF diagnosis and their caregivers (CFF, 2019b). The next step is to discover more specific influences that contribute to the elevated rates of

anxiety and depression in caregivers (and patients) and how to best help this population with emotional wellness whilst awaiting a cure.

Purpose Statement

The purpose of this study was to understand the lived experiences of caregivers raising children with CF in relation to rates of anxiety, depression, and quality of life, as they experience a burden of treatment care that few parents go through and understand. This study aimed to explore the connection between lived experiences, context, and systemic influences contributing to the elevated rates of psychological symptoms within the CF community from the caregiver's perspective. The combination of numerical and experiential data in this mixed methods study provided a unique opportunity to better understand the caregiver's quality of life, psychosocial functioning of the child (as reported by the caregiver), managing the child's health complications, familial and social struggles, and other obstacles these caregivers face on a daily basis.

In conducting this study, there was an opportunity to identify and understand the contributing systemic and biopsychosocial factors to caregiver prevalence of anxiety, depression, and chronic illness care stressors based on the mixed methods approach. The study highlighted the individualized barriers to caregivers in seeking and receiving their own mental health care and the relation to increased rates of anxiety and depression and lower quality of life scores.

Rationale/Significance of Study

The information gathered by this mixed methods study will be used to supplement the training various mental health clinicians receive on how to deliver higher quality

therapeutic services to this demographic in and outside the CF Care Clinics. It will also contribute to the gap in literature where personalized, experiential data has been lacking in research. It has been documented that individuals with CF, and their caregivers, experience a greater burden from the child's illness and treatment than other populations; therefore, obtaining a better understanding of the psychological stressors that plague this demographic throughout the lifespan while maintaining mental health is of the utmost importance (Garcia et al., 2018; Smith et al., 2016). This study also attempted to recruit a higher number of male caregiver participants to better understand their involvement and treatment burden, as they are generally missed in CF research (Shardonofsky, Cesario, Fredland, Landrum, Hiatt, et al., 2019b). Although the number was not as high as expected, almost 10% of the study participants were male caregivers. Due to the online nature of this study, the goal of recruiting more male participants seemed feasible as a majority of children are brought to CF clinic appointments by their mothers (Cronly et al., 2019).

Theoretical Foundation

This study was guided by the family systems genetic illness model (FSGI), a secondary version of Rolland's family systems illness (FSI) model, updated to include the biological, genetic component and influence within the family system (Rolland & Williams, 2005). The original FSI model focused on family functioning in the realm of chronic conditions after clinical onset, whereas the FSGI model incorporates the component of time (before, during, and after diagnosis) and how families adjust, adapt,

and cope throughout various phases of illness and disorder throughout the lifespan for all family members, not just the diagnosed individual (Rolland & Williams, 2005).

In 1977, George Engel proposed a biopsychosocial approach to medicine with a move from the familiar "traditional" biomedical model to a more integrated and humanistic approach that takes into account not only biological factors, but psychological factors and the social context of the diagnosed individual (Engel, 1977). Rolland expands upon Engel's seminal work by discussing the need for an integrative approach that is defined in terms of family systems theory where the family unit (all caregiving and/or family members) are the focus of care (Rolland, 1994). Both the FSI and FSGI models are integrative, collaborative, systemic, developmental, family-centered models that are prevention-oriented and resilience based (Rolland, 2018).

The FSGI model was selected due to the holistic and collaborative approach between the diagnosed individual, family caregivers and loved ones, and the medical care team; all of which are vital components of families within the CF community. The desire to focus specifically on the FSGI rather than FSI was based on the genetic component and inclusion of the dimension of time throughout the lifespan of the FSGI. Regarding the genetic component of CF: this is a type of illness that is different from many other chronic conditions with varying degrees of severity and the possibility that an individual may be a carrier, but not have CF. Within the FSGI model, CF is specifically listed in the genomic typology due to the genetic carrier aspect (Rolland & Williams, 2005). Although individuals may not have CF, when they are carriers there is a risk their children will

have CF, which creates a psychological component to be aware of within the family system and from a multigenerational perspective (Rolland & Williams, 2005).

Research Questions

This mixed methods research study was based on one overall research question combining qualitative and quantitative components. This study sought to understand the lived experiences of caregivers raising a child with a CF diagnosis when considering the quality of life of that family, the psychosocial functioning of the child (as reported by the caregiver), the experience of managing the child's health complications, the family's social struggles, and other obstacles the caregiver may face on a daily basis. The study also sought to identify contributing factors of overall caregiver prevalence of anxiety, depression, and chronic illness care stress from a quality of life standpoint. These factors were assessed by utilizing the following instruments: General Anxiety Disorder-7 (GAD-7), Patient Health Questionnaire-9 (PHQ-9), Caregiver Quality of Life Cystic Fibrosis (CQOLCF), and the Cystic Fibrosis Questionnaire-Revised (CFQ-R).

Research Question One: How do female caregivers and male caregivers raising a child with a cystic fibrosis diagnosis differ in rates of anxiety, depression, and quality of life?

Research Question Two: How does a referent child's CFTR genetic mutation and CF severity relate to caregiver rates of anxiety, depression, and quality of life?

Research Question Three: How does the referent child's age and sex affect caregiver anxiety, depression, and quality of life when that child has a cystic fibrosis diagnosis?

Research Question Four: How does access to CF Care Clinics affect caregiver anxiety, depression, and quality of life?

Research Question Five: How do biopsychological factors contribute to elevated rates of anxiety and depression, and lower rates of quality of life for caregivers who are raising a child with a cystic fibrosis diagnosis?

Please see Table 1, which provides an outline for key terms and their definitions used within the study and this research.

Table 1

Table of Definitions

Term	Definition
Cystic Fibrosis	A life-shortening, chronic and genetic illness affecting the mucous membranes and digestive system (CFF, 2014b).
Child with CF	A child between the ages of 0-18 with a cystic fibrosis diagnosis under the care and monitoring of a primary caregiver.
CF Diagnosis Severity	Mild, Moderate, Severe, Awaiting Lung Transplant, Received Lung Transplant *As diagnosed by the child's primary medical physician (Farrell et al., 2017).
CF Caregiver	An individual primarily in charge of caring for a child with cystic fibrosis including but not limited to: administering treatments and medications, taking to doctor and hospital visits, appointments, and surgeries; responsible for the general health and well-being of the child; having legal authority to make psychological and physical/medical health decisions on behalf of the minor child; over the age of 21; and identified as mother, father, stepparent, grandparent, aunt, uncle, foster parent, guardian.
Chronic Disease	A chronic condition requiring continued medical treatment which limits activities of daily living, lasting one year or more (Centers for Disease Control and Prevention [CDC], 2019).
Mental Health	"Mental health is a dynamic state of internal equilibrium which enables individuals to use their abilities in harmony with universal values of society. Basic cognitive and social skills; ability to recognize, express and modulate one's own emotions, as well as empathize with others; flexibility and ability to cope with adverse life events and function in social roles; and harmonious relationship between body and mind represent important components of mental health which con- tribute, to varying degrees, to the state of internal equilibrium" (Galderisi et al., 2015, p. 231–232).
Mental Health Professional	Psychologists (PhD/PsyD), Master's level Counselors (LPC), Clinicians (LCDC), Therapists, Clinical Social Workers (LICSW, LCSW, LMSW), Psychiatrists (MD/DO), Psychiatric or Mental Health Nurse Practitioners (MHNP), Primary Care Physicians (PCP), Family Nurse Practitioners (FNP), Psychiatric Pharmacists, Pastoral Counselors, Bachelors level Social Workers, and Certified Peer Specialists (NAMI, 2019).

Assumptions

1. Parents and caregivers were honest in their reports.
2. The child was diagnosed with cystic fibrosis by a medical professional.
3. The participants in this study were truly caregivers for a child diagnosed with CF.

Inclusion Criteria

1. Participants were caregiving adults, 21 and older, who were caring for a child, from birth to 18 years, with a CF diagnosis.
2. Caregivers included mothers, fathers, stepparents or any other adult responsible for the full-time care and support of a child from birth to18 years with a CF diagnosis. Alternate caregivers included grandparents, aunts, uncles, foster parents, guardians, who had legal authority to make medical and mental health decisions for the child.
3. Participant caregivers were responsible for the care of the CF diagnosed child at least 50% of the time.
    a. Male caregivers were especially encouraged to participate.
    b. Participants were accepted if they lived in the United States.
    c. Participants were required to be able to read, write, and understand English.

Delimitations

1. Caregivers of children with CF over the age of 18 were excluded from participation as this portion of the demographic is typically able to provide their own care, except under extreme circumstances or a progressed case of CF.

Caregivers were required to have a child with a CF diagnosis under the age of 18 in their primary care. A participant could have a child with CF who was older than the age of 18, but in order to meet study requirements, they must also have had a child under the age of 18 with a CF diagnosis.

2. Caregivers of children with other genetic, life-shortening illness were excluded due to the nature of the treatments and medical attention this population requires in order to slow disease progression and survival. The primary diagnosis of the child had to be CF to be eligible to participate in the study. If a child had CF-related diagnoses such as CF-related diabetes (CFRD), CF-related asthma, CF-related bone disease, and CF-related liver disease, then, caregivers were still able to participate in the study.
3. Individuals who did not read, write, and understand English were excluded from the study as all components of the survey were in English.

Summary

This chapter discussed the prevalence and impact of a cystic fibrosis diagnosis on individual and relational functioning from a physical, mental, and emotional viewpoint. CF is the number one genetic, life-shortening, chronic illness of Caucasians (CFF, 2014a). The daily survival requirements for CF patients ranges from routine and emergency doctor's appointments and hospitalizations, vest treatments, and multiple medications and enzymes throughout the day. Social activities, educational opportunities, and familial obligations are burdened and require special considerations due to the child's survival needs taking priority, which take a toll on the primary caregiver of the child. The

purpose of this research was to understand the lived experiences of caregivers of children with CF. Many individuals with CF, and caregivers of children with CF, acknowledge the lack of time for individual, couple, or family therapy as daily physical treatments are required focus to ensure the survival and comfort of the child. The rationale for the study was to understand the special circumstances of these families so mental health professionals may effectively and efficiently incorporate enhanced psychological services into their protocol of treatments.

A brief review of literature discussed historical and recent data pertaining to the CF population and the impact this life-shortening disease has on rates of anxiety, depression, and quality of life for individuals and caregivers raising a child with CF. This chapter identified the problem and purpose statements, rationale and significance of the study, theory guiding this research, research questions, tables of definitions, assumptions of the study, and delimitations of participants for the study.

CHAPTER II

REVIEW OF LITERATURE

Introduction

CF is a rare, genetic, life-shortening disease, primarily affecting Caucasians, with approximately 1,000 new cases being diagnosed annually (CFF, 2014b). CF was once considered a childhood disease, however, advances in medical technology over the past 30 years have increased the likelihood of survival into adulthood. CF treatments are determined based on the severity of the disease, genetic mutations, and are established on an individual basis. Daily treatment times range from two to four hours including pancreatic enzymes, medications, inhalers, nebulizers, and chest physiotherapy. For families with a loved one with a CF diagnosis, treatments become part of life incorporated into the daily routine; however, nothing tends to be spontaneous as caregivers of children with CF have reported their lives revolve around these daily treatments. The focus of CF can take its toll physically and emotionally on both the diagnosed individual as well as their loved ones. Recent studies indicated elevated levels of depression and anxiety and lower levels of quality of life of CF patients as well as their caregivers.

This chapter will provide an in-depth discussion of CF and its history, its genetic component, and the ways in which the disease effects the family system. The purpose of this study was to explore the experiences of caregivers raising children with a CF diagnosis and the relationship between rates of anxiety, depression, and quality of life.

This chapter presents an overview of these key variables, the role of CF, and recent data supporting the findings of increased rates of anxiety and depression and decreased rates of quality of life through the lens of the FSGI Model developed in the 1980s by John S. Rolland.

Cystic Fibrosis: Genetics and Statistics

According to the National Heart, Lung, and Blood Institute (NHLBI; 2013b), CF is a disease of the secretory glands that causes a thick, sticky mucus to buildup in the lungs, pancreas, liver, intestines, sinuses, and sex organs. CF is a rare, recessive disease passed down when both parents have a mutation of the cystic fibrosis transmembrane conductance regulator (CFTR) protein (CFF, 2014a).

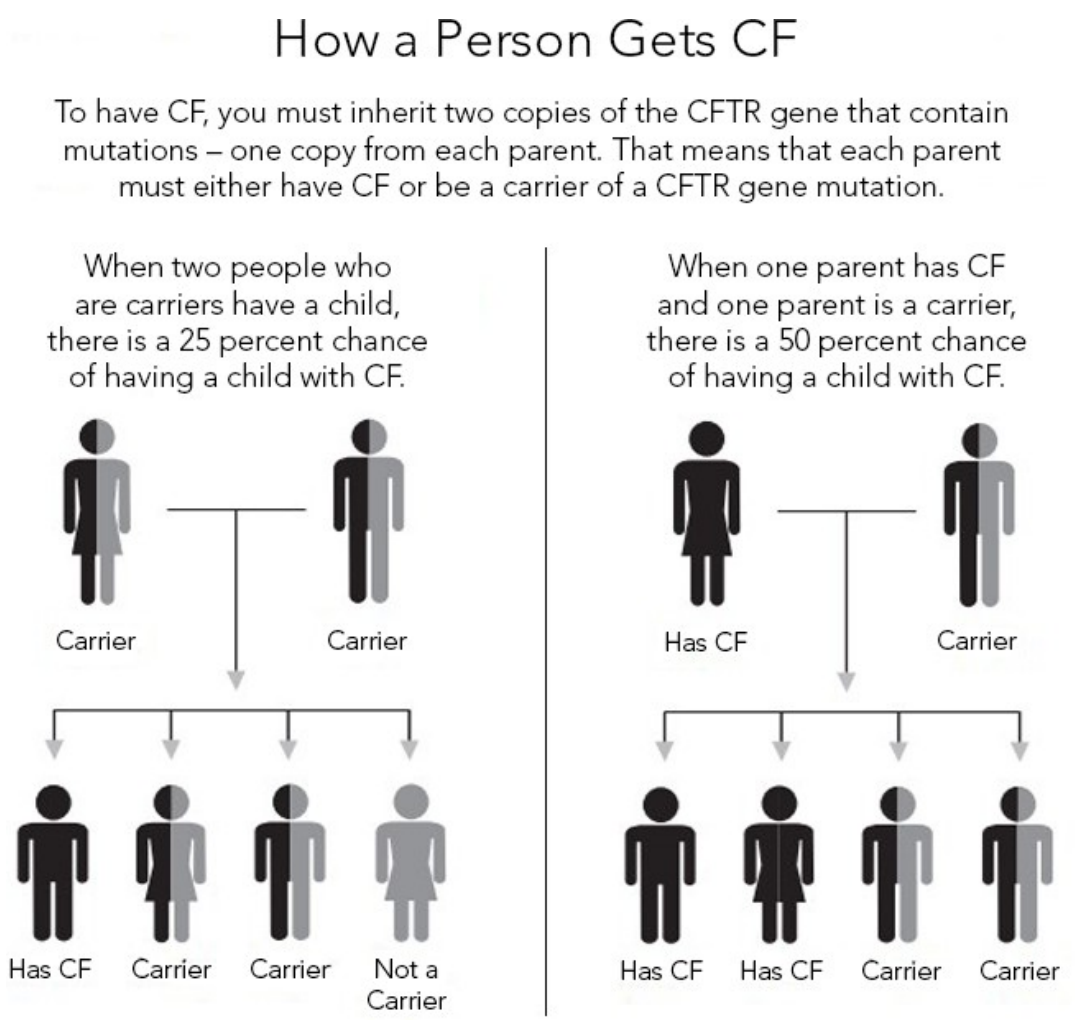
The CFTR protein regulates the flow of salt and fluid within the cells of the body and helps to maintain the balance of salt and water on internal surfaces such as the lungs. When the CFTR gene is mutated, the protein may not work well, have insufficient quantities, or may not be produced at all. When the CFTR protein is not working properly, the body is unable to maintain the balance of salt and fluids and a component of salt, called chloride, becomes trapped within the cells. As chloride blocks the water from hydrating the cell's surfaces, the airways and organs become dehydrated and a thick, sticky mucus forms that leads to the complications specific to CF, including difficulty breathing, airway infections, lung failure, poor digestion, and reproductive problems (CFF, 2014b). Individuals who do not have CF normally produce mucus in the body that is thin and slippery and acts as a lubricant to protect various linings, organs, and tissues; whereas a person with CF produces thick and sticky mucus that will clog airways that can

lead to inflammation, infections, and permanent damage of the lungs (National Library of Medicine, 2015).

Every individual has two copies of the CFTR gene; CF carriers are individuals who have one copy of the mutated gene and one normal gene (CFF, 2019a). When both parents have one mutated and one normal gene, they may have a child with cystic fibrosis (CFF, 2014a). When two CF carriers have a child, there is a 50% chance (1 in 2) that the child will be a carrier but will not have CF; a 25% chance (1 in 4) the child will have CF; and a 25% chance (1 in 4) the child will not have CF nor be a carrier (CFF, 2019a). Individuals with CF may also pass CF gene mutation copies to their children. There is a 50% chance (1 in 2) the child will be a carrier but will not have CF and a 50% chance (1 in 2) the child will have CF (CFF, 2019a). Please see infographic "*How a Person Gets CF"* from the CFF (2019a) in Figure 1. Approximately 10 million people in the United States are CF carriers.

Figure 1

How a Person Gets CF



Note. Adapted from *How a Person Gets CF* by CFF (2019a; https://www.cff.org/What-is-CF/Testing/Carrier-Testing-for-Cystic-Fibrosis/. In the public domain.

There are approximately 70,000 to 100,000 cases of CF worldwide; the estimated number is indefinite due to the lack of appropriate healthcare methods of tracking and CF specific databases in developing and underdeveloped countries (CFWW, 2019). Additionally, the number varies based on inadequate healthcare being provided in

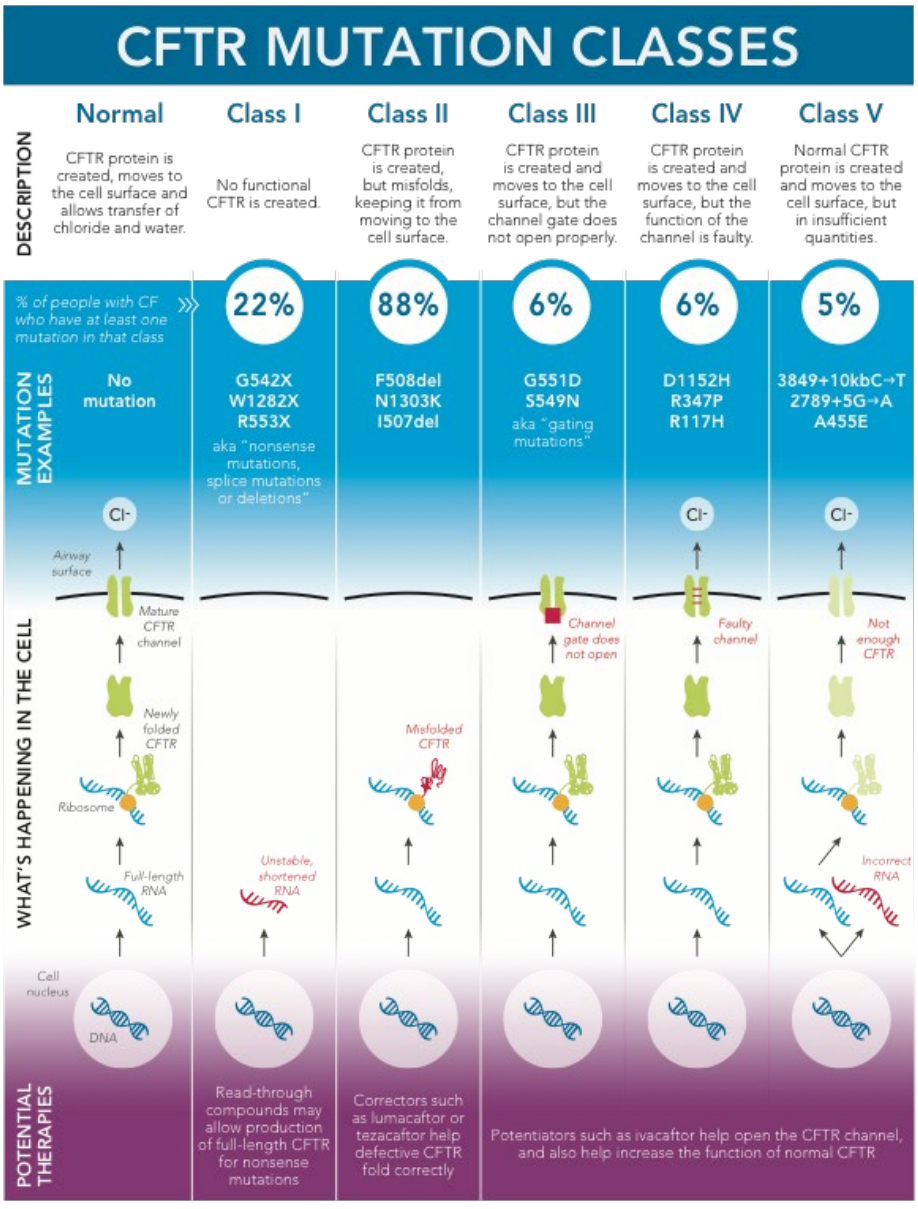
countries such as India, El Salvador, and Bulgaria, where CF is thought to be extremely rare and the life expectancy decreases to 15 or younger (CFWW, 2019).

Classes of Cystic Fibrosis

While overall characteristics of CF are similar, the difficulty treating the physical problems lie in the various classes of CFTR mutations, making it difficult to pinpoint specific treatments for each affected individual (CFF, 2017a). There are over 1700 variations of the CFTR gene and five classes of mutations, the most common of which is the F508del mutation (CFF, 2017a). Approximately two-thirds of all worldwide CF mutations are the F508del mutation, leaving the remaining one-third with great mutational heterogeneity (Asper Biogene, 2020). Other common CFTR gene mutations include G542TER, G551D, and nonsense mutations which account for approximately 10% of CF cases worldwide (Asper Biogene, 2020). Nonsense mutations are non-functioning proteins that have been created due to premature completion of CFTR production caused by a faulty stop signal (CFF, 2017b). Mutations are not always able to be neatly classified as one specific class and are grouped based on the problems caused within the production of the CFTR protein (CFF, 2017b). Class one are protein production mutations, class two are protein processing mutations; class three are gating mutations; class four are conduction mutations; and class five are insufficient protein mutations (CFF, 2017a; 2017b). Please see infographic “CFTR Mutation Classes” from the CFF (2017a) in Figure 2.

Figure 2

CFTR Mutation Classes



Note. Adapted from *CFTR Mutation Classes* by CFF (2017a; https://www.cff.org/What-is-CF/Genetics/Know-Your-CFTR-Mutations-Infographic.pdf). In the public domain.

Health Risks for People Diagnosed with Cystic Fibrosis

According to the NHLBI (2013c), individuals diagnosed with CF are at a higher risk for respiratory infections due to abnormal mucus production. These abnormal mucus productions decrease their ability to break down food and absorb vital nutrients in the pancreas. Individuals with CF may have difficulty breathing, shortness of breath, chronic cough, abdominal pain, delayed growth, developmental delays and be more prone to infection (Mayo Clinic, 2012). Individuals tend to develop gastrointestinal complications, often suffer from dehydration, have increased heart rates, fatigue, weakness, decreased blood pressure, heat stroke, and are at a higher risk of developing diabetes, osteoporosis, and osteopenia, than individuals without CF (NHLBI, 2013c). Reproductive systems are affected in both men and women with cystic fibrosis: men are born without vas deferens, the tube that delivers sperm from the testes to the penis, and are infertile, while women have difficulty conceiving due to mucus blocking the cervix or other complications related to the disease (NHLBI, 2013c).

Treatments for Cystic Fibrosis

Treatments include, but are not limited to: oral antibiotics to prevent and/or treat lung infections; anti-inflammatory medicines to reduce swelling and keep airways clear; mucus-thinning drugs to break up mucus to be orally expelled; oral pancreatic enzymes to assist the digestive tract in absorbing essential nutrients; bronchodilators to relax muscles around airways and keep them open; and chest physical therapy with chest clappers, inflatable vests, and breathing devices (NHLBI, 2013a). Medical advances have also led to the introduction of CFTR modulator medications that are designed to correct

malfunctioning proteins; however, these gene therapy medications are only available to individuals with specific mutations (CFF, 2014b). Individuals who are physically able may go through pulmonary rehabilitation achieved through exercise, nutritional counseling, energy conserving practices, breathing strategies, and counseling services. Many individuals require various surgeries such as nasal polyp removal, oxygen therapy, endoscopy and lavage, lung transplants, feeding tubes, and bowel surgery (Mayo Clinic, 2012).

CF is a disease that is all encompassing and is more than simply taking medication to minimize symptoms. It is a high treatment burden, time-consuming disease with treatments taking up to four hours a day (Sawicki et al., 2009). With multiple daily medications, airway clearance treatments, chest physical therapy, frequent hospital stays, CF-related complications such as diabetes, asthma, and liver disease, and an ongoing battle with sterilizing medical devices and maintaining an environment that is as germ-free as possible to decrease potential infections and illness, the emotional and mental well-being of individuals with CF and their caregivers naturally comes into question.

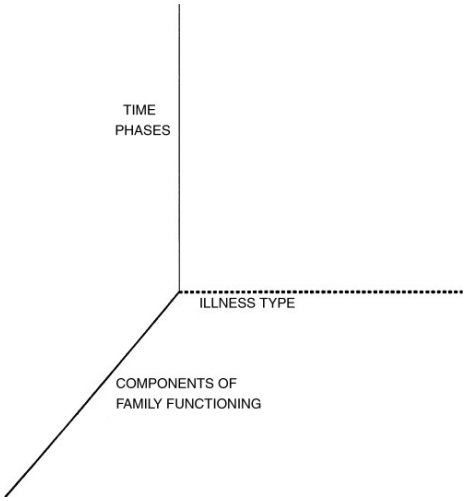
Family Systems Genetic Illness Model Theory

Rolland (1994) posited where chronic and life-threatening conditions are present, a systemic, biopsychosocial model addressing the psychosocial needs of all family members and friends is needed. As briefly mentioned in chapter one, the FSGI is a collaborative and integrative family systems approach that is prevention oriented and resilience-based across the lifespan (Rolland, 2018; see Figure 3). This model assesses illness type and components of family functioning throughout time phases while

emphasizing interactions of illness with belief systems, multigenerational patterns of coping with illness, grief, and loss, and individual and family development (Rolland & Williams, 2005), all of which is vital to understand the meaning and experiences of families raising a child with a CF diagnosis.

Figure 3

The Family Systems Illness Model



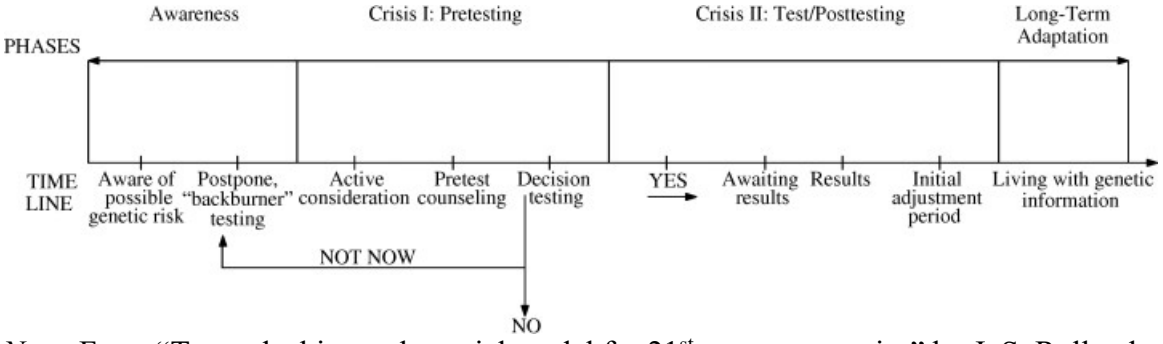
Note. From "Toward a biopsychosocial model for 21st century genetics" by J. S. Rolland & J. K. Williams, 2005, *Family Process, 44*(1), 3-24, p. 6. (https://doi.org/10.1111/j.1545-5300.2005.00039.x) Copyright 2005 by the FPI, Inc.

The standard of care for CF diagnosis is generally by the age of 2, with a majority of 64% of diagnoses being completed with newborn screening (NBS; Farrell et al., 2017). However, there are individuals who opt not to complete NBS and diagnosing non-screened individuals can be challenging due to symptom severity, age of onset, and CFTR dysfunction (Farrell et al., 2017). This data is a diagnostic test and does not take

into consideration the psychological well-being of the parents, their life cycle phase, or the parental and/or familial development. This is where FSGI may be applied, as the theory was established to help families better understand and master their lives with the illness as part of the family. Within this model, families are given the opportunity to better understand how the disease unfolds throughout various phases (Rolland, 1994). FSGI identifies patterns of emotional, practical, and psychosocial demands to expect throughout various developmental stages for both the diagnosed individual and their families (Rolland, 1994; see Figure 4). It allows family members to learn how to become a functional family unit over the course of disease progression through open communication, shared meaning, and understanding (Rolland, 1994). FSGI offers the ability for increased awareness and appreciation of changes of both the ill individual and family members as each progress throughout personal family life cycles (Rolland, 1994). Finally, this model helps both families and clinicians better grasp the multigenerational influences, values, and beliefs that inform their health struggles, meaning of the experiences, and how time effects their dynamic (Rolland, 1994).

Figure 4

Nonsymptomatic Time Phases of Genomic Disorders



Note. From "Toward a biopsychosocial model for 21st century genetics" by J. S. Rolland & J. K. Williams, 2005, *Family Process, 44*(1), 3-24, p. 12. (https://doi.org/10.1111/j.1545-5300.2005.00039.x) Copyright 2005 by the FPI, Inc.

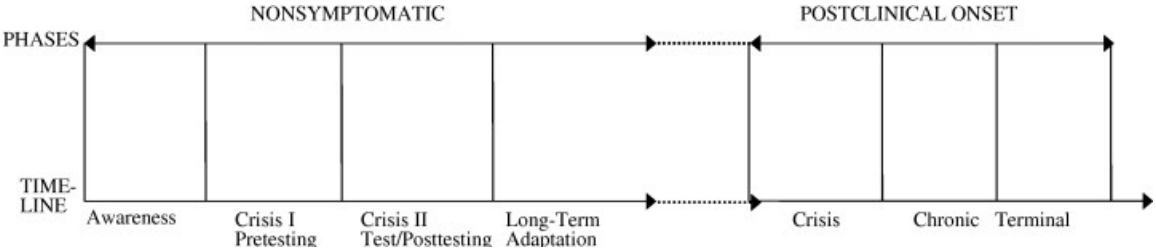
It has been briefly mentioned that CF is a time-consuming disease with multiple daily treatments, medications, and doctor's appointments taking up to 4 hours per day (Sawicki et al., 2009). For caregivers raising a child with CF, until their child is at an age where they are able to monitor and take responsibility for their daily treatments, this is a high time burden for the parent to ensure completion. How is the caregiver's physical and emotional health affected if they are a single parent, have multiple children, work multiple jobs, and/or lack financial resources and social support? The FSGI is a family-centered model that helps to understand how the disease impacts all family members and relationships and is designed to examine these relationships between individuals and family dynamics with genomic disorders (Rolland & Williams, 2005).

Utilizing a model that takes into consideration the dimension of time allows for contemplation of disease onset, chronic conditions and/or acute symptoms and illness, outcome, incapacitation, and terminal phases (Rolland & Williams, 2005). CF is a disease

that can be ruled out with genetic testing completed by potential parents before they have children (carrier testing), during pregnancy through amniocentesis (prenatal testing), with newborn screening, and post NBS screening when a child develops CF like symptoms (diagnosis/prognosis). The FSGI offers a psychosocial framework for genomic disorders such as CF and provides the opportunity to uncover support interventions when considering: (1) timing of clinical onset: child/adolescent (birth-20 years), early/mid adulthood childrearing (20-60), and later life (60+); (2) the likelihood of development (parental CF gene carriers); (3) clinical severity; and (4) if medical interventions are available to alter onset and/or progression (Rolland & Williams, 2005; see Figure 5). The FSGI model focuses on diseases that have carrier testing, predictive, and presymptomatic testing currently available, or that may come available in the future (Rolland & Williams, 2005). CF is a disease that can be identified through carrier testing, with a small window of predictability based on parental carrier status. The FSGI incorporates a genetic component to the FSI model while being based on the same three principles and provides a model for families to better understand their own genetic risk, explore the shared familial meanings of health, illness, and loss, and how this information influences family functioning over time (Rolland & Williams, 2005).

Figure 5

Time Phases of Genomic Disorders



Note. From "Toward a biopsychosocial model for 21st century genetics" by J. S. Rolland & J. K. Williams, 2005, *Family Process, 44*(1), 3-24, p. 17. (https://doi.org/10.1111/j.1545-5300.2005.00039.x) Copyright 2005 by the FPI, Inc.

In the FSGI, there is a level of uncertainty that hovers above the genomic typology variables (Rolland & Williams, 2005). Please see Table 2 that provides the components of the FSGI Model and identifies the model dimensions, applications to CF, and the application to genetic risk; adapted from Daly (2015).

Table 2

Dimensions of Family Systems Genetic Illness Model

FSGI model dimensions	Application to CF	Application to genetic risk
Illness Type	Type of CF genetic mutation	Type of genetic risk: are both parents' carriers?
	Mode of onset	Likelihood of child developing CF
	Phase of life cycle at CF diagnosis for the child and caregiver	Phase of life cycle when receiving test results (parent and child)
	Availability of therapy	Availability of preventative or therapeutic options
	Course and severity of CF	Expected severity of disease
Time Phases of CF	Awareness of CF carrier status	Awareness of family risk
	Diagnostic testing (child and caregiver)	Information seeking
	Diagnosis (prenatal, NBS, symptomatic prognosis)	Testing decisions
	Treatment decisions	Adoption of screening (carrier and prenatal testing)
	Treatment phases	Active monitoring of symptoms
	Adaptation to chronic and acute phases	Adaptation to genetic risk phase
	Terminal phase	Outcomes of genetic risk – continued throughout childbearing years of CF carrier
Family Systems Variables (Components of Family Functioning)	Family experience with CF	Family experience with genetic risk
	Coping with CF skills	Coping with genetic risk skills
	Caregiving skills	Family support structures
	Family values and beliefs about CF	Family values and beliefs about genetics
	Communication patterns about CF	Communication patterns about genetic risk
	Quality of familial relationships	Quality of familial relationships
	Adapting new family roles	Adoption of new risk identity (will children bear the same psychological burden of parents)?

Note. Adapted from "A family-centered model for sharing genetic risk" by M. B. Daly 2015, *Journal of Law, Medicine & Ethics, 43*(3), 546 (https:doi-

org.ezp.twu.edu/10.1111/jlme.12297). Copyright 2015 American Society of Law, Medicine, and Ethics.

Psychological Screening and Mental Health Assessment

Besier and Goldbeck (2011) highlighted the possibility that psychological struggles may not be presented and/or discussed during routine clinic visits and identify the need for regular mental health screening and referrals when appropriate. Cronly et al. (2019) identified the need for greater study into the mental health and psychological well-being of parent caregivers as this is a chronic-life shortening disease with an extremely high treatment burden. The research also recommends online screening of caregivers to minimize the possibility of missing paternal involvement.

Anxiety and Depression in the United States

Anxiety and depression are the two most commonly diagnosed mental illnesses in the US, with over 40 and 17 million annual depression and anxiety diagnoses, respectively (NIMH, 2019). The Anxiety and Depression Association of America (2018) identified approximately 18% of the total population as experiencing an anxiety disorder in any given year, making it the most commonly diagnosed mental illness. The National Alliance on Mental Illness (NAMI, 2017) state there are various anxiety disorders, but they all share common characteristics including persistent, excessive fear or worry in situations that are not threatening but can interfere with daily functioning in relationships, school, work, and social interactions. Nearly 50% of individuals diagnosed with depression are also diagnosed with anxiety (Anxiety and Depression Association of America, 2018).

According to the NIMH (2019), data from the 2017 National Survey on Drug Use and Health identify over 17.3 million adults over the age of 18 to have experienced a major depressive episode in the past year. Females (8.7%) have a higher prevalence of depression than males (5.3%), with the highest prevalence based on the adult age group 18–25 (13.1%). Additionally, approximately 3.2 million adolescents ages 12–17 experienced a major depressive episode in the past year, representing 13.3% of the total population for this age bracket. Again, prevalence was higher for female adolescents (20%) when compared to males (6.8%). To qualify as a major depressive episode, the *Diagnostic and Statistical Manual of Mental Disorders* (5th ed; DSM-5) is defined as, "a period of at least two weeks when a person experienced a depressed mood or loss of interest or pleasure in daily activities, and had a majority of specified symptoms, such as problems with sleep, eating, energy, concentration, or self-worth" (NIMH, 2018, para. 2). The survey did not make exclusions based on medications, substance abuse, or symptoms caused by medical illness.

Anxiety and depression are commonly associated with cystic fibrosis amongst children, teens and adolescents, and adults as well as parent caregivers (Besier et al., 2011; Cruz et al., 2009; Driscoll et al., 2010; Hodgkinson & Lester, 2002; Quittner et al., 2008; Quittner et al., 2014). Child health related quality of life is directly negatively associated with their parent's depression and anxiety scores; parents who report higher levels of anxiety and depression also report lower quality of life in their children (Cronly et al., 2019). It has been hypothesized/documented that higher disease severity tends to correlate highly with lower quality of life scores (Boling et al., 2003). Besier and

Goldbeck (2011) conducted a study to evaluate life satisfaction across the lifespan in four different age groups (12–20, 21–30, 31–40, 40–64 years) of German individuals with CF and found elevated symptoms of anxiety and/or depression as having a strong negative association with life satisfaction as they were used as indicators of psychological well-being. Besier and Goldbeck (2011) established a connection with mental health being negatively affected and having the possibility of negatively influencing adherence to medical treatment across the lifespan. Depression is also a known risk factor to poor treatment adherence within chronic conditions (DiMatteo et al., 2000).

Anxiety and Depression in Caregivers Raising a Child with Cystic Fibrosis

There are limited studies assessing caregiver rates of anxiety and depression for those raising a child with CF (Driscoll et al., 2009). A majority of mental health related data have been targeted at the diagnosed population, thus published data for their caregivers remains limited (Cronly et al., 2019). However, what the minimal research shows is that rates vary from 12–33% of male caregiver experiences of anxiety (Quittner et al., 2014).

The International Depression and Anxiety Epidemiological Study (TIDES) established elevated prevalence of psychological symptoms in adults and adolescents with CF as well as parent caregivers (Quittner et al., 2014). This study was a major effort to identify and understand the association of psychological symptomology within the CF community. The authors recruited a large sample of 6,088 patients and 4,102 parent participants across 154 CF centers throughout the US and Europe and found elevated rates of depression as follows: adolescents (10%), adults (19%), mothers (37%), and

fathers (31%); and anxiety: adolescents (22%), adults (32%), mothers (48%), and fathers (36%) within the sample population. Additional significant correlations were found, and the result of TIDES was a recommendation for annual psychological screening for patients and their parent caregivers (Quittner et al., 2014).

Cronly et al. (2019) found in an Irish sample that 38% of participant caregivers had elevated anxiety scores; 12% had elevated depression scores; 31.7% reported they had experienced mental or emotional problems within the previous 12 months; and over 22% reported they sought professional help for their distress. Mothers reported higher anxiety scores than that of participant fathers. Individuals who were single and unable to work had higher rates of anxiety and depression than those who were in a relationship and working part- or full-time. A correlation between child hospitalization and pulmonary lung function within the past year and caregiver depression was also noted. Child health related quality of life (HRQOL) for ages 6–13, also had significantly negatively correlations associated with caregiver rates of anxiety and depression. For parents of children ages 14–17 there was a significant negative correlation associated with child emotional functioning and caregiver depression score.

Father Involvement

A majority of CF research reflects an assumption that caregiving roles are delegated to mothers, focusing on the maternal mental health of female caregivers; as such suggesting a lack of paternal involvement or father caregiver burden within the family unit (Shardonofsky, Cesario, Fredland, Landrum, Hiatt, et al., 2019b). Cronly et al. (2019) reported difficulty in recruiting father participants as a majority of children

were brought to clinic appointments by their mothers; therefore, fathers have a lower likelihood of being screened for psychological struggles and have a small chance of being identified (and provided appropriate referrals) as needing mental health care services. Historically, a majority of caregiver research is centered around female involvement due to traditional gender roles, maternal attachment, and accessibility; there is a documented lack of father involvement in CF research (Cronly et al., 2019; Shardonofsky, Cesario, Fredland, & Landrum, 2019a). This study sought to increase the number of male participants in order to understand their perspective and identify a wider range of experiential biopsychosocial factors contributing to elevated rates of anxiety and depression as well as lower rates of quality of life for the CF caregiver demographic. Including male participants in this current study allowed them the opportunity to have a greater voice within the CF research.

Previous research conducted through CF center clinic visits targeted the caregiver who brought the child to their appointment, generally mothers, and missed the father demographic due to the nature of sampling and recruitment (Cronly et al., 2019). The choice to include father caregivers, rather than exclude them in this study was with the intention of having increased father representation, better understanding the role of fathers in CF care, father-specific struggles, as well as father rates of quality of life, anxiety, and depression. By conducting this study and collecting participant data online, it removed the outside constraints of already stressful quarterly CF clinic visits (generally completed by mothers), minimizing the likelihood that male caregivers would be overlooked or missed, thus there was ample opportunity for male caregivers of children

with CF to participate in the current study.

Previous research found fathers identified themes of financial strain, feeling overwhelmed, experiencing altered family dynamics, isolation, actively seeking resources, and feeling hopeful (Shardonofsky, Cesario, Fredland, Landrum, Hiatt, et al., 2019b). Different types of normal, when things get difficult, and decision-making complexity were themes which emerged based on routine management of treatments, dealing with symptoms, and battling CF related decline from a case study of one mother raising a child with CF (Glascoe & Smith, 2011). Hodgkinson and Lester (2002) found emergent themes of feeling stuck in the middle, coping with changing identity, and the burden of responsibility in a study of mothers of children with CF. Finally, Jessup et al. (2018) found five main themes with two subthemes in a small Australian, rural study including: daily care: a family affair, accessing expert care (subtheme), the CF circle, control versus collaboration: seeking mutual trust, family-centered care: seeking inclusion, the team who grows with you (subtheme), and future projections.

An Unprecedented World-Wide Pandemic

When the current study was first proposed in March of 2020, the United States was in the beginning stages of planned shut downs, limiting travel, and closing businesses and employment positions that were deemed "nonessential" to help slow the spread of the novel Coronavirus, also known as Severe Acute Respiratory Syndrome Coronavirus-2 (SARS-COV-2), COVID-19, and in slang terms, "The Rona." We were, and still are, collectively experiencing a worldwide global pandemic due to a virus that originated in Wuhan, China. As of March 2021, there have been over 123 million cases

of coronavirus (WorldOmeter, 2021). What was originally thought to be a self-limiting airborne virus that appeared to be an inflammatory respiratory illness similar to the flu or pneumonia, has evolved into a potentially life-threatening multisystem inflammatory disease involving multiple organs and bodily systems (Hatmi, 2021). Symptoms of coronavirus range from those similar to the common cold or flu including stomach upset, fever, headache, cough, and shortness of breath, to nasal congestion, rapid breathing, muscle soreness, and acute respiratory distress syndrome. Individuals with underlying conditions such as autoimmune diseases, hypertension, respiratory illnesses, diabetes, and those who are obese, or smoke cigarettes are at a higher risk of experiencing complex comorbidities and complications related to COVID. Individuals with cystic fibrosis are considered in the higher risk category of developing more serious complications if they end up with SARS-COV-2, a single positive-strand RNA virus that is primarily contracted from person-to-person contact via airborne droplets or surface contamination transfer (CFF, 2020a; Hatmi, 2021). Those within the cystic fibrosis community were already accustomed to keeping up with the challenges of maintaining as "germ-free" living environments as possible through good hygiene and sanitizing, and some may also have been used to wearing a mask in order to prevent the spread of airborne germs and bacterial infection when at clinic, hospital visits, and out and about running errands.

When the study went live in July of 2020, it appeared to be the height of the pandemic with many worldwide areas still under high levels of lockdown, curfews, and quarantine ordinances to follow. In hindsight, including more questions regarding the pandemic specifically and how it has affected this population as a whole would have

benefited the current study greatly. Undoubtedly, coronavirus influenced a majority of participants with reported struggles ranging from being geographically displaced, losing employment or being overworked (due to the nature of employment), and financial strain. Another main concern stated was for the physical and mental health safety of the participant's children, especially their child with CF, as well as the participants. Many of the study's participants discussed how the lack of social support and interactions, physical and emotional isolation, changing governmental policies regarding rules, regulations, lockdowns, and quarantine contributed to their feelings of overwhelm, anxiety, depression, and perceived quality of life. Daily life transitioned almost overnight from in person outings to virtually everything moving online. Participants had to navigate online/virtual learning and/or homeschool changes as well as working remotely, which many parents discussed to be stressors within their families. The strain of the pandemic was felt by all participants in one way or another. The influence of the coronavirus pandemic is discussed in Chapter 4 and was present in each of the experiential questions, except for the fifth, as that question was focused on gene modulating medications.

Summary

Continued medical advances have led to increased life expectancy with individuals diagnosed with CF. What was once known as a deadly childhood and adolescent disease with low-life expectancy has transitioned to adults currently leading full lives into their 40s, 50s, and 60s. It is important to address taking care of those diagnosed with CF throughout the lifespan from a holistic and systemic perspective. In recent years, there has been a shift within the CFF as it recognized the importance of

identifying the emotional and psychological toll of the illness on the individuals diagnosed. Increases in research to understand the psychological needs of this population have been growing with a great emphasis on anxiety, depression, quality of life, relationships, and life satisfaction for individuals with CF. More recent data from the TIDES study indicated that not only do individuals with CF have elevated rates of anxiety and depression, along with diminished rates of quality of life, relationships, and life satisfaction, their caregivers do as well. The unexpected stressors of an unprecedented global pandemic have also led to heightened physical health concerns for those within the high-risk category such as individuals with CF. The short and long-term physical, emotional, social, and financial impacts of the coronavirus for the world's population are still unknown.

CHAPTER III

METHODOLOGY

Introduction

To expand and explore the impact of childhood CF on families, a mixed-methods explanatory study to identify and understand the experiences of caregivers raising children with a CF diagnosis and the relationship between elevated rates of anxiety and depression, and diminished rates of quality of life was proposed. The combination of both qualitative and quantitative data was paramount to this study as the goal was to understand the psychosocial influences impacting the relationship in rates of anxiety, depression, and quality of life in caregivers raising a child with CF. When qualitative and quantitative data are linked numbers and words are both needed in order to understand the world of the participants (Miles et al., 2014). When participants' truths, meaning, subjective realities, and experiences are a topic of research exploration, it is reasonable to incorporate a phenomenological element into the study (Miles et al., 2014). Although a mixed methods approach is more time consuming, there are many benefits to this type of research including hypothesis generation and testing, triangulation, development of appropriate research tools and strategies, complementarity, completeness, and resolution of puzzle finding (Miles et al., 2014). The combination of quantitative and qualitative data for the study can be found in the statistical analysis section of Chapter 4.

In this chapter, the study methods presented include sample recruitment, identification of the study sample, sample, protection of human subjects, research

questions and hypotheses, caregiver and child demographic questionnaires and variable tables of measurement, quantitative instrument information, qualitative survey questions, procedures, study components and participant survey instructions, incentives, phenomenological genre background, and statistical analysis selection of multivariate analysis of variance (MANOVA) data analysis methods.

Sample Recruitment

The United States population of individuals diagnosed with CF totals approximately 30,000, with over half (15,000) being younger than 18 years old (CFF, 2018). After consultation with the Texas Woman's University (TWU) Center for Research Design and Analysis (CRDA), an estimated sample size of 300 adult caregiver participants was set for recruitment from adult caregivers within CF communities across the US. Recruitment was conducted via an online cross-sectional, non-probability, purposive (initial approach) and snowball (secondary approach) sample of United States residents over the age of 21. Potential participant family groups were contacted online via Facebook and Instagram utilizing the following hashtags:

> #cf, #cysticfibrosis, #cysticfibrosisawareness, #cysticfibrosislife, #CFwarrior, #cfer, #cfsucks, #fucf, #cfirl, #curecf, #justbreathe, #lovetobreathe, #clairesplacefoundation, #chronicillness, #invisibleillness, #spoonies, #65roses, #smellthe65roses, #fivefeetapart, #lungtransplant, #doublelungtransplant, #newlungs, #organdonation, #donatelife, #parenting, #raisinggentlemen, #raisingladies, #letthembe, #theartofslowliving, #cfmom, #cfdad, and #cfparent.

Identification of Study Sample

Prior to beginning the current study, at the request of my major advisor, I contacted the administrators of online Facebook community groups with group membership combined over 27,000 people to make a preliminary request to post recruitment information about this proposed study within their groups. One particular group on Facebook has over 7,000 members and is limited only to mothers and fathers of children with a CF diagnosis. Although I was unable to join this group as I am not a caregiver raising a child with CF, a group administrator happily agreed to share the study recruitment within the group privately. Other group administrators gave their verbal approval in September 2019 pending Institutional Review Board (IRB) application approval (see Appendix A). Upon IRB approval, follow-up requests to post recruitment data in the online Facebook groups was submitted to the group administrators who provided preliminary approval. A couple in California, my acquaintances, agreed to connect with their local CF chapters who had expressed interest and support in sharing the study on their social media outlets. I am also connected with the CFF Community Innovators group and was able to share the study within a privately-run group on Facebook. All posts were sent to the public via Facebook and Instagram with the opportunity for others to share on their personal social media as well, thus further expanding the reach.

I shared the recruitment advertisement (see Appendix B) on my own social media channels. I also contacted www.Cystic-fibrosis.com and they shared on their website and social media channels my request seeking participants. The PsychData survey was

located at https://cystic-fibrosis.com/spotlight/raising-child-survey/ but has now been closed. The survey link was available online 24 hours a day, 7 days a week during the study timeframe. Potential participants went to the survey link to express interest in the study from a time, date, and location which fit their schedule and availability. The online survey was closed when I reached 297 participants due to academic time constraints. Participants were also asked if they were interested in future research related to CF and mental health care.

Sample

The participants in this study were primarily mothers and fathers from 21 to 69 years who were raising a child diagnosed with CF ranging in age from birth to age 18. Participant inclusion criteria required caregivers to hold legal authority to make medical and mental health decisions for the child, to be able to read, understand, and write in English, and reside in the United States. Male and female respondents within the same household who were caring for the same child at the time of the study were permitted to participate. Separated and/or divorced caregivers, living in separate households, who had 50% physical custody of the diagnosed child were also permitted to participate. Sample descriptive statistics for parent participants and their referent child are provided in chapter four.

Protection of Human Subjects

I followed TWU IRB requirements for quantitative and qualitative online research. Participants in this study completed an anonymous online questionnaire in which they were asked general demographic data such as age, gender, race, marital

status, and education. Participants were asked questions about their daily time commitment in assisting their child with treatments, adherence to medical protocol, and implications of these routines on their quality of life and psychosocial (daily mental, emotional, physical, and social abilities) functioning. Participants were asked questions regarding how raising a child with a chronic illness diagnosis has affected relationships with family and friends. Participants were asked to complete a series of questionnaires and assessments to quantify individual levels of anxiety, depression, and quality of life.

The risks were as follows: fatigue, emotional exhaustion, time lost to complete the survey, emotional discomfort and/or distress at sharing information about the illness of their child, loss of time, and potential loss of confidentiality due to the online nature of the study. Confidentiality was protected to the extent that is allowed by law. Participation was completely voluntary. Participants were able to withdraw at any time without penalty. Participants were given a list of national behavioral health providers as the study reached a national audience of parent caregivers.

Research Questions and Hypotheses

Research Question One: How does age and sex of caregivers relate to rates of anxiety, depression, and quality of life?

H_a1: Female caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression), and lower scores of CQOLCF (quality of life) when compared to scores of male caregivers.

Research Question Two: How does a referent child's CFTR genetic mutation and CF severity relate to caregiver rates of anxiety, depression, and quality of life?

H_a2: Caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression) and lowers scores on the CQOLCF (quality of life) when compared to their child's CFTR genetic mutation and CF severity; caregivers of children with greater CF severity and rarer genetic mutations will have higher anxiety and depression scores and lower quality of life scores.

Research Question Three: How does the referent child's age and sex affect caregiver anxiety, depression, and quality of life?

H_a3: Caregivers with infants and female adolescent children will have higher scores on the GAD-7 (anxiety), PHQ-9 (depression) and lower scores on the CQOLCF (quality of life).

Research Question Four: How does access to CF Clinic affect caregiver anxiety, depression, and quality of life?

H_a4: Scores of caregivers on the GAD-7 (anxiety), PHQ-9 (depression) and CQOLCF (quality of life) will be negatively impacted by distance from CF clinic.

Research Question Five (qualitative): How do biopsychological factors contribute to elevated rates of anxiety and depression, and lower rates of quality of life for caregivers who are raising a child with a cystic fibrosis diagnosis?

Variables and Instruments

The following section identifies the quantitative questionnaires and qualitative survey questions used within the study. This section also provides variable tables showcasing definitions, type of measurement, and level of measurement for both caregiver participants and their child with cystic fibrosis.

Caregiver Demographic Questionnaire

Participant caregivers completed a demographic questionnaire providing information for themselves including gender, age, race, ethnicity, marital status, level of education, employment status, insurance coverage, number and age of children, number and age of children with CF, child education status, nearest CF Care Center, anxiety, depression, other mental health diagnosis, mental health treatment, and psychotropic medication. The caregiver quantitative variables can be found in Table 3 and the caregiver demographic questionnaire may be found in Appendix C.

Caregiver Quantitative Variables

Table 3

Caregiver Variables: Definition, Type, Level of Measurement

Variable	Type of variable	Level of measurement	Definition	Assessment
Gender	Independent	Nominal	Biological Sex M/F	Self-report
Age	Independent	Ratio	Chronological Age	Self-report
Caregiver Quality of Life	Dependent	Interval	The quality of life as reported by the caregiver	Caregiver Quality of Life-Cystic Fibrosis (CQOLCF)
Anxiety	Dependent	Interval	Level of anxiety reported by caregiver	General Anxiety Disorder-7 (GAD-7)
Depression	Dependent	Interval	Level of depression reported by caregiver	Patient Health Questionnaire-9 (PHQ-9)

Note. Limitations: all self-report data may not be 100% accurate and/or reliable.

Child Demographic Questionnaire

Participant caregivers completed a child demographic medical history questionnaire providing information about their CF diagnosis and health related information. Demographic questions include: age of child; age of CF diagnosis; gender; CF gene mutations: FEV_1, commonly referred to as pulmonary function test (PFT); BMI; weight; CF severity; lung transplant waiting list; previous lung transplant; co-morbid diagnosis; tune-up within past 6 months; nasal surgery within past 6 months; positive cultures for Pseudomonas aeruginosa, Mycobacterium abscessus, and Burkholderia cenopacia; anxiety; depression; other mental health diagnosis; mental health treatment; and use of psychotropic medication. The child quantitative variables can be found in Table 4 and the child demographic questionnaire may be found in Appendix D.

Child Quantitative Variables

Table 4

Child Variables: Definition, Type, Level of Measurement

Variable	Type of variable	Level of measurement	Definition	Assessment
Age	Independent	Ratio	Chronological age	Self-report
Gender	Independent	Nominal	Biological sex male/female	Self-report
CF Gene Mutation 1	Independent	Nominal	Category of CF	Self-report
CF Gene Mutation 2	Independent	Nominal	Category of CF	Self-report
CF Severity	Independent	Ordinal	Mild, Moderate, Severe (as dx by PCP)	Self-report
FEV_1	Independent	Ratio	Forced expiratory volume in 1 second, a measure of pulmonary function (Boling et al., 2003)	Self-report
PFT	Independent	Ratio	Pulmonary function test	Self-report
Child Quality of Life	Dependent	Interval	The child's quality of life as perceived and reported by the caregiver.	Cystic Fibrosis Questionnaire-Revised (CFQ-R) Self-report

Note. Limitations: all self-report; physical health data may not be 100% accurate.

Quantitative Surveys

The quantitative assessment surveys used within the study, rationale for their inclusion, and brief examples of survey questions are discussed below. The surveys may be found in the appendices as follows: GAD-7 (see Appendix E); PHQ-9 (see Appendix F); CQOLCF Scale (see Appendix G); and CFQ-R (see Appendix H).

The General Anxiety Disorder-7

The GAD-7 is a 7-item, 4-point Likert-type scale self-report assessment with criteria in which general anxiety disorders are based from the Diagnostic and Statistics Manual-IV (DSM-IV), and had the ability to establish symptom severity as well as anxiety diagnosis (Spitzer et al., 2006). Individuals were asked to answer statements reviewing the past 2 weeks; items are scaled from "0" (*not at all*) to "3" (*nearly every day*), to questions such as "Feeling, nervous, anxious or on edge," "Trouble relaxing," and "Feeling afraid as if something awful might happen." Scores range from 0–21, with cutoff points of 5, 10, and 15 to represent mild, moderate, and severe anxiety, respectively. The GAD-7 has excellent test-retest reliability (interclass correlation = 0.83), internal reliability with a Cronbach's α of 0.92, and good construct, criterion, factorial, and validity (Spitzer et al., 2006). The GAD-7 is a brief, concise measure that is time efficient, making it a dual-purpose instrument to assess anxiety diagnosis and anxiety severity.

The Patient Health Questionnaire-9

The PHQ-9 is a 9-item, 4-point Likert-type scale self-report assessment with criteria in which depressive disorders are based from the DSM-IV and had the ability to establish symptom severity as well as depressive diagnosis (Kroenke et al., 2001). Individuals were asked to answer statements considering the past two weeks; items are scaled from "0" (*not at all*) to "3" (*nearly every day*), to statements such as: "Little interest or pleasure in doing things," "poor appetite or overeating," and "Thoughts that you would be better off dead or of hurting yourself in some way." Scores range from 0–27, with cutoff points of 5, 10, 15, and 20 to represent mild, moderate, moderately severe, and severe

depression, respectively. The PHQ-9 has excellent test-retest reliability as well as internal reliability with a Cronbach's α of 0.89, and rates high of construct, criterion, and validity (Kroenke et al., 2001). The PHQ-9 is a brief, concise measure that is time efficient, making it a dual-purpose instrument to assess depression diagnosis and symptom severity.

The Caregiver Quality of Life Cystic Fibrosis Scale

The CQOLCF scale is a 35-item, 5-point Likert-type scale self-report assessment to assess the quality of life for caregivers of individuals with cystic fibrosis (Boling et al., 2003). The CQOLCF was created to assess the most relevant health-related needs and concerns of the cystic fibrosis population and is based off the Caregiver Quality of Life-Cancer scale. Individuals were asked to answer statements considering the past seven days. Items are scaled from "0" (*not at all*) to "4" (*very much*), to statements, such as: "I worry about the impact my loved one's illness has had on my other children or other family members;" "I get support from my friends and neighbors;" and "The responsibility I have for my loved one's care at home is overwhelming." Scores range from 0–140 with higher scores representing better quality of life. The CQOLCF has a split-half correlation coefficient of 0.862 ($p < 0.01$), excellent internal reliability with a Cronbach's α of 0.909, and has been found to be valid, reliable, and consistent measure assessing quality of life of caregivers utilizing the CF disease-specific scale.

The Cystic Fibrosis Questionnaire-Revised

The Cystic Fibrosis Questionnaire was originally developed in 1997 in France and was revised and updated for use in English in the United States by Dr. Alexandra Quittner (Habib et al., 2015). The CFQ-R measures 9 disease-specific quality of life

dimensions including physical functioning, energy/well-being, emotions, social limitations, role, embarrassment, body image, eating disturbances, and treatment burden (Quittner et al., 2005). The questionnaire is a 35–50 item, 4-point Likert-type scale interview (ages 6–11) and self-report formats (ages 12 & 13, and 14+, and parent caregiver of children (6–13) used to measure quality of life of children, adolescents, and adults with CF. In each format, individuals were asked to answer statements considering the past 2 weeks. Scores range from 0–100 with higher scores representing better health.

In the online study, if participant caregivers selected the 14 and older CFQ-R questionnaire, they were asked to coordinate with their child to obtain their feedback. However, there was a possible likelihood that parents felt as if they were unable to accurately complete the questionnaire on behalf of their child. Parents were asked to select a drop-down menu item after the completion of this questionnaire to identify: (1) if their child completed the assessment independently (self-report format 12–13, and 14+); (2) if the parent interviewed the child and recorded their responses (interviewer format 6–11); or (3) if the parent answered the questionnaire on behalf of their child without their input (parent/caregiver 6–13, self-report 12–13, and 14+). It was my goal to obtain as accurate and robust data as possible and I recognize this aspect of the study is a potential limitation, which will be discussed in greater detail in the limitations section.

The Interviewer Format for ages 6–11, is composed of 35, 4-point Likert-type items scaled from 1–4. Question scoring was dependent upon the category in which the respondent answered with possible selection groupings being as follows: “1” (*very true,*

always, very true), "2" (*somewhat true, often, mostly true*), "3" (*somewhat false, sometimes, somewhat true*) and "4" (*very false, never, not true at all*) to statements such as:

(16) "you had to stop fun activities to do your treatments"

(23) "you felt left out"

(32) "you woke up during the night because you were coughing"

The Self-report Format for ages 12 & 13, is composed of 35, 4-point Likert-type items scaled from 1–4. Question scoring was dependent upon the category in which the respondent answered with possible selection groupings being as follows: "1" (*very true, always*), "2" (*mostly true, often*), "3" (*somewhat true, sometimes*) and "4" (*not at all true, never*) to statements such as:

(5) "You were able to participate in sports that you enjoy (e.g., swimming, soccer, dancing or others)

(12) "You had trouble falling asleep"

(29) "You thought you were physically different from others your age"

The Self-report Format for ages 14 and above, is composed of 50, 4-point Likert-type items scaled from 1–4. Question scoring was dependent upon the category in which the respondent answered with possible selection groupings being as follows: "1" (*a lot of difficulty, always, very true, a great deal*), "2" (*some difficulty, often, somewhat true, somewhat*), "3" (*a little difficulty, sometimes, somewhat false, a little*) and "4" (*no difficulty, never, very false, not at all*) to statements such as:

(16) "how much time do you spend each day on your treatments?"

"1" a lot, "2" some, "3" a little, "4" not very much

(23) “I feel comfortable discussing my illness with others”

(32) “I feel healthy”

(50) “Have you had eating problems?”

The Self-report Format for Parent/Caregivers of children ages 6–13, is composed of 35, 4-point Likert-type items scaled from 1–4. Question scoring was dependent upon the category in which the respondent answered with possible selection groupings being as follows: “1” (*a lot of difficulty, always, very true, a great deal*), “2” (*some difficulty, often, somewhat true, somewhat*), “3” (*a little difficulty, sometimes, somewhat false, a little*) and “4” (*no difficulty, never, very false, not at all*) to statements such as:

(5) “To what extent has your child had difficulty climbing several flights of stairs”

(13) “Indicate how often your child was absent or late for school because of his/her illness or treatments”

(24) “Indicate the extent to which each sentence is true or false for your child: My child lives a normal life”

(36) “Please indicate how your child has been feeling during the past two weeks: My child had to cough up mucus”

The CFQ-R is the most commonly used patient reported outcome measure for individuals with CF and their parents (Alpern et al., 2015; Habib et al., 2015). The CFQ-R consistently demonstrates good sensitivity to change, validity, and reliability (Quittner et al., 2005). Cronbach α was used to calculate internal consistency in scale level reliability with reliability coefficients ranging from $r = 0.18$ to 0.94; test-retest reliability has intraclass correlations from 0.45 to 0.90 for most domains (Quittner et al., 2005).

Pulmonary function, BMI, age, and CF disease severity relationships were used to determine validity of the measure utilizing a multivariate analysis of variance; Hotelling $T^2 = 0.44$, $F[22,384] = 3.84$, $p < 001$ (Quittner et al., 2005). Similar domains on the CFQ and SF-36 were used to determine convergent validity including physical ($r = 0.81$, $p < 0.01$), health perceptions/general health ($r = 0.79$, $p < 0.01$), vitality ($r = 0.84$, $p < 0.01$), role/role-physical ($r = 0.73$, $p < 0.01$), emotional functioning/mental health ($r = 0.74$, $p < 0.01$), and social ($r = 0.57$, $p < 0.01$; Quittner et al., 2005). Relationships between domains on the CFQ and SF-36 that did not measure similarly were used to determine discriminant validity; example general health and mental health ($rs = 0.19$ to 0.42; Quittner et al., 2005).

Qualitative Survey Questionnaire

The goal of the open-ended qualitative questions was to uncover and describe perspectives held by participant caregivers regarding their history and experience of raising a child with cystic fibrosis, and their perceptions of how the illness impacts their lives (Marshall & Rossman, 2016). The interview questions were pre-determined, open-ended, and semi-structured in order to allow the participants to identify how they view the topic and their emotions, feelings, and thoughts associated (Marshall & Rossman, 2016).

To obtain a more encompassing experiential viewpoint to incorporate with quantitative data, qualitative questions for this study were open-ended to allow participants to interpret them based on their own family experiences and provide input based on the meaning they associated with their lived experiences with CF, while keeping in mind

themes found in past research. The Qualitative Survey Questionnaire may be found in Appendix I.

Procedure

Consent Form and Data Collection

This online study was voluntary and available through PsychData from July 6, 2020, to August 6, 2020; during this time 297 parents raising children with CF began the survey. Links to the PsychData survey were included on all social media platforms sharing in order to direct participants directly to the study. The survey link was available online 24 hours a day, 7 days a week during the study timeframe. Potential participants went to the survey link to express interest in the study from a time, date, and location which fit their schedule and availability.

The first page of the online survey, participants were provided an Informed Consent (see Appendix J) document which provided information for criteria and limits of participation, outlined the study risks (loss of confidentiality, loss of time, fatigue, emotional discomfort and/or distress), with additional information regarding the purpose and intent of the research, and time expectations (35–50 minutes). Participants were advised that should they become fatigued or need to take a break, they may stop and take a break at any time, but the survey did not allow for saving and returning. Participants were prompted to self-confirm they qualified for the study and understood the informed consent; they were notified that if they continued to the next page, they were giving their consent to be a voluntary participant in this study.

The second page of the online survey prompted participants to answer the following statement in a Yes/No format: “Please check this box if you are aware that your child's other parent caregiver, in the same household, is also completing this study. Participants were then provided Participant Survey Instructions (see Appendix K) along with a breakdown of the process including study components and time commitment, which may be found in Table 5.

Table 5

Study Components

Study component	Number of questions	Approximate time (minutes)
Reading about the Study and Informed Consent	N/A	5–10
Participant Demographic Questionnaire	22	3–5
Child Demographic Questionnaire	27	3–5
Qualitative Question One	1	3–5
General Anxiety Disorder-7 (GAD-7)	7	1–2
Qualitative Question Two	1	3–5
Patient Health Questionnaire-9 (PHQ-9)	9	1–3
Qualitative Question Three	1	3–5
Caregiver Quality of Life Cystic Fibrosis (CQOLCF) Scale	35	5–10
Qualitative Question Four	1	3–5
Cystic Fibrosis Questionnaire-Revised (CFQ-R)	Varies by age grouping; no more than 45	15

Incentives

No incentives were provided. Participants had the comfort of knowing they were contributing to the overall gap in literature for the CF Community. Participants who elected to receive a report of the results summary will be notified via email.

Data Analysis Methods

Genre: A Phenomenological Approach

This study utilized a phenomenological approach to facilitate a better understanding of the lives of caregivers as they explored, described, and analyzed the meaning of their experiences (Marshall & Rossman, 2016). This method provided an opportunity for caregivers to give meaning to the health and illness stories of their families, while bridging the gap in literature to connect the rich qualitative data to explore the quantitative, numerical data identifying rates of caregiver anxiety, depression, and quality of life. According to Patton (2002), phenomenology is "how they perceive it, describe it, feel about it, judge it, remember it, make sense of it, and talk about it with others" (p. 104).

A phenomenological framework seeks to understand the essence of a collective group's experiences, where the focus is on the life as it is lived via the participant (Marshall & Rossman, 2016). This approach allowed the qualitative portion of this mixed methods study to provide holism, richness, and thick descriptions (Miles et al., 2014) of the emotional, systemic, and familial meaning, challenges, and resilience of families with a child with a cystic fibrosis diagnosis. For a phenomenological qualitative approach to be effective, researchers are encouraged to "bracket" their own preconceptions and values and be led completely by the meaning structure associated with the topic by the participant (Gergen, 2014).

Qualitative Analysis

The purpose of the online qualitative portion of the study was to answer the fifth research question, "How do biopsychological factors contribute to elevated rates of anxiety and depression, and lower rates of quality of life for caregivers raising a child with cystic fibrosis?" Participants answered five experiential, open-ended questions that sought to uncover their lived experiences from their own perspective and reality from a phenomenological approach in order to bridge the connection to the quantitative components of the study. The Qualitative Survey Questionnaire may be found in Appendix I. Participants answered questions relating to barriers to seeking and receiving individual mental health care, their experiences with anxiety and depression, how they view cystic fibrosis as impacting their quality of life, as well as their perspective on the continued advances in CFTR gene modulator medication therapy availability and what this may mean for their child. Participants were provided a 1,000-word limit in the online survey to voice their experiences and were given little direction or guidance in order to not skew the results or lead the participant in any way.

Experiential Coding

In order to become familiar with the qualitative data, I downloaded all the data from PsychData into a Microsoft Excel file in order to have an easily accessible version of all responses to separate questions in one place to identify common feelings, phrases, or themes. The justification for this process was to take advantage of the ability to openly code the data while searching for common patterns and key ideas that may be shared between participants (Marshall & Rossman, 2016). As the survey had three age grouped

surveys within the survey, I merged all the data into a master file, and completed an overall read through of all participant responses once to familiarize myself with the data and to obtain a general understanding of the participants experiences. I then collapsed the age grouped responses into one tab for each question, from one to five and completed a second reading where I looked for key phrases and patterns for each question. The third read I narrowed down the key phrases and patterns, identified categories across each of the experiential questions, and then I grouped them into themes.

Next, I created two identical copies of the master coding list to prepare to send to two doctoral level research assistants. Second level coders were used to confirm my first impressions of emergent key words, phrases, themes, and patterns in order to minimize researcher bias and for intercoder reliability and consistency (Marshall & Rossman, 2016). I confirmed there was no overlap in participant responses being reviewed by each coder by sorting their coding sheets differently. Each coding sheet provided every fourth participant response for their review and consideration due to the high number of participant responses. Both coders independently confirmed the emergent categories, patterns, and initial impressions I identified and provided additional feedback regarding inclusion of additional information and potentially creating new categories and/or themes. The breakdown of responses reviewed by coder per question can be seen in Table 6.

Table 6

Second-Coder Review Breakdown

Question number	Responses reviewed per coder (*n*)	Total responses reviewed (*n*)	Total participant responses (*n*)	Responses reviewed by second coders (%)
One	22	44	82	53.69
Two	20	40	81	49.38
Three	15	30	57	52.63
Four	14	28	58	48.28
Five	13	26	56	46.43

My major advisor served as a third level coder to further confirm themes, categories, and patterns identified by myself and the second level coders. At the time of writing, it was proposed that every 20th participant's qualitative responses be read and coded by the second level coders, however, as mentioned above, the random sampling number changed. From this point, my major advisor went through and sorted through the qualitative data as well. She read through all responses once to obtain a general feel for participant responses and experiences and then completed a second read-through to identify and confirm any additional key words, phrases, emergent patterns, and/or themes.

As I continued to read through the response of the participants to the first three rounds of coding, I had a feeling that I couldn't put into words. Thankfully one of my second level coders was able to identify the language I was seeking and eloquently stated an overarching theme she had found present throughout her coding process, which were the ideas of natural skepticism and cautious optimism. During the coding process, I also noticed the theme of mental health stigma continuing to present itself in various formats,

and second coders confirmed. My major advisor expressed hesitation and I went back through my coding and confirmed with my second coders on two separate occasions; first to confirm the theme as a whole, next to confirm the presence of the four types of stigma. Both coders reported "overwhelmingly" and "wholeheartedly" that the responses of participants fell into one, if not multiples, of the mental health stigma categories. As participants discussed the contributing factors and their lived experiences relating to each qualitative question, five emergent themes were identified: (1) The CF Priority, (2) Resources and Support, (3) Pandemic Amplification of Distress, (4) Natural Skepticism and Cautious Optimism, and (5) Barriers to Seeking and Receiving Mental Health Services. Where present, subthemes are included in the analysis, all of which are discussed in Chapter 4.

Quantitative Statistical Analysis

Descriptive statistics were used to characterize caregiver and child demographic data, including means, standard deviation, and frequencies (Babbie, 2012). Caregiver descriptive statistics may be found in Table 8, child descriptive statistics may be found in Table 10, and together in Appendix L. A thorough review of the descriptive statistics may be found in chapter four. In order to best understand the relationships and differences between the variables within this study, MANOVA was used to answer all research questions to minimize familywise error rate (Huck, 2012). The MANOVA allowed us to understand the effect of caregiver sex, child's CFTR mutation, CF severity, and CF clinic access (independent variables) on caregiver scores of anxiety, depression, and quality of life (dependent variables; Babbie, 2012). In order to account for the established

relationship between the dependent variables, MANOVA was used to identify how groups (male versus female caregivers) differ from one another while allowing for the simultaneous examination of the independent variables (Babbie, 2012). Multivariate regression was originally going to be used to answer research question three, to understand the impact of child age and sex (independent variables) on caregiver rates of anxiety, depression, and quality of life (Babbie, 2012). However, after further consideration, this analysis would have required three multiple regressions to be computed which could have led to an increase in type II error; therefore, a MANOVA was used for this research question as well. A breakdown of the testing for each research question can be found in Table 7.

Table 7

Research Question Statistical Analysis

Research question	Independent variable	IV variable type	Test
One	Caregiver status Sex: M/F	Nominal	MANOVA
Two	CFTR Mutation CF Severity	Nominal	MANOVA
Three	Child Age Child Sex	Variable one: interval Variable two: nominal	MANOVA
Four	CF Clinic Distance	Ordinal	MANOVA

Note. The dependent variables for each research question are caregiver rates of anxiety, depression, and quality of life; all are interval variables.

Blending Mixed Methods Results

The patterns, categories, and themes that emerged from the data were used to better understand the quantitative data obtained from participant scores on the study's assessments. Combining quantitative and qualitative methods for the study benefits the

gap in literature by adding experiential context from the participants with numerical data, which helped to identify the factors that influenced participant's daily biopsychosocial functioning. The potential to understand the needs of this population by combining quantitative and qualitative analysis is fundamental to better serving the mental health and psychological struggles of this demographic.

By providing completeness, a more comprehensive understanding of psychosocial factors and the relationship of rates of anxiety, depression, and quality of life was possible (Bryman, 2006). With complementarity, elevated rates of anxiety and depression and lower rates of quality of life were discussed throughout the first two chapters. What was lacking is the connection to the numerical data to better understand the meaning of the lived experiences of this demographic. Through this process where quantitative data may show a weakness (unknown origin of causation or correlation of numerical relationships), the qualitative data provided a bridge of connection to understand these relationships and vice versa (Miles et al., 2014).

Hypothesis generation and testing: the aim of this research was to better understand the psychosocial influences and relationship of quantitative data of participants in order to increase the dialogue of needed interventions and mental health care for this demographic. The qualitative data obtained helped to inform the cause and effect relationships within the CF community (Miles et al., 2014). Development of appropriate research tools and strategies: data from previous research guided the formulation of the current study by identifying rates of anxiety, depression, and quality of life, and introducing qualitative themes which emerged in prior research to inform the

open-ended survey questions here. The qualitative data helped to construct questions for quantitative data and the assessments being utilized (Miles et al., 2014). Triangulation of data through qualitative inquiry and multiple quantitative assessment measures provided the opportunity to have greater validity when they were found to support the research questions and conclusions (Bryman, 2006).

Resolution of puzzle finding in this research occurred when the qualitative data was able to provide personalized meaning of lived experiences of participants which helped to better understand the quantitative numerical data (Miles et al., 2014). Participant qualitative responses were used to explore the quantitative findings through concurrent mixed methods where the qualitative data was paralleling or gathering the quantitative data at the same time. In order to best integrate the findings, the qualitative analysis was performed first to minimize quantitative data biases. Once the qualitative data had been coded, the quantitative statistical analysis were computed in the form of MANOVA. The quantitative data was then used to come back to inform the qualitative data that had already been coded for emergent themes and patterns.

Summary

A mixed-methods approach to data collection has many benefits including hypothesis generation, testing, triangulation, complementarity, completeness, and the ability to find missing pieces in puzzle finding (Miles et al., 2014). This study utilized an online cross-sectional, non-probability, purposive (initial) and snowball (secondary) approach in recruiting 297 American caregivers over 21, raising a child with a CF diagnosis. Recruitment was done completely online via Facebook, Instagram, and word

of mouth. The study sought to increase paternal input to better understand the needs of both mothers and fathers raising children with CF. This study sought to understand the lived experiences of caregivers raising a child with CF and the quality of life of the caregivers as well as the psychosocial functioning of the child (as reported by the caregiver).

Participants completed quantitative assessment measures including PHQ-9, GAD-7, CQOLCF, and CFQ-R. Participants answered five qualitative open-ended questions to explore their lived experiences and personalized meaning, and the impact of CF on their daily family life. The research genre for this study was a phenomenological approach to facilitate a better understanding of the meaning of the experiences of participants (Marshall & Rossman, 2016). MANOVAs were used to test the effects and relationships between multiple independent and dependent variables (Huck, 2012). Qualitative and quantitative data were analyzed together to zoom out and better understand the psychological needs of a population riddled with physical health struggles. Tables of variable definition, type, and level of measurement were also provided in this chapter.

CHAPTER IV

RESULTS

Introduction

The purpose of this mixed-methods study was to explore and understand the lived experiences of caregivers raising children with CF in relation to their rates of anxiety, depression, and quality of life, as their experiences as a family within the CF community differs greatly from caregivers of children with other chronic illness and/or the well population. This study aimed to identify the connection between the systemic influences contributing to the elevated rates of psychological symptoms within the CF community from the caregiver's perspective of their self-reported lived experiences and individual context. Utilizing a mixed methods approach of collecting both qualitative and quantitative data provided an incomparable opportunity to thoroughly investigate how systemic biopsychosocial factors influence caregiver rates of anxiety, depression, and quality of life while taking into consideration familial, financial, cultural, and social strengths and struggles, managing daily CF care and treatment, as well as routine and typical day-to-day stressors.

This study sought to answer the following research questions:

1. How do female caregivers and male caregivers raising a child with a cystic fibrosis diagnosis differ in rates of anxiety, depression, and quality of life? The alternative hypothesis stated is female caregivers will have higher scores of

anxiety, depression, and lower scores of quality of life when compared to scores of male caregivers.

2. How does a referent child's CFTR genetic mutation and CF severity relate to caregiver rates of anxiety, depression, and quality of life? The alternative hypothesis states caregivers will have higher scores of anxiety and depression, and lower scores of quality of life when compared to their child's CFTR genetic mutation and CF severity; caregivers of children with greater CF severity and rarer genetic mutations will have higher scores of anxiety and depression and lower quality of life scores.
3. How does the referent child's age and sex affect caregiver anxiety, depression, and quality of life when that child has a cystic fibrosis diagnosis? The alternative hypothesis states caregivers with infants and female adolescent children will have higher scores of anxiety and depression, and lower scores of quality of life.
4. How does access to CF Clinics affect caregiver anxiety, depression, and quality of life? The alternative hypothesis states that scores of anxiety, depression, and quality of life for caregivers will be negatively impacted by the distance from CF clinic.
5. How do biopsychological factors contribute to elevated rates of anxiety and depression, and lower rates of quality of life for caregivers who are raising a child with a cystic fibrosis diagnosis? Research question five does not have an alternative hypothesis as was experiential data collected through open-ended questions in qualitative format.

Quantitative Findings

Establishing the Final Quantitative Data Set

The initial data set had 297 participants; however, when the data set was reviewed, 142 participants had missing, invalid, and/or incomplete quantitative data, or did not fit the inclusion criteria. These cases were removed, resulting in a usable sample of 155 parents raising children with CF. All 155 participants completed demographic and child-related data, with a few exceptions. Some participants did not answer demographic specific questions and would skip over them and continue to the next portion. In order to obtain the most holistic view of the data as possible, the decision was made to include this information even if the sample size for a specific question was not $N = 155$; the discrepancies are noted and discussed later in this chapter.

Procedures for Descriptive Statistics and Statistical Analyses

Descriptive statistics were conducted on key parent and child variables as well as the three dependent variables of parent anxiety, depression, and quality of life. Simple frequency distribution was used to identify participant and their referent child's demographic information including age, gender, CF severity, CFTR gene mutation, etc. Measures of central tendency including the mean and median were used to quantify the measures of anxiety, depression, and quality of life in the GAD-7, PHQ-9, and CQOLCF, respectively. Parametric MANOVA testing was utilized to determine relationships between variable groups to answer the first four research questions. Measures of variability including range, standard deviation, and variance are presented in the data analysis of the research question section of this chapter. Participant descriptive

information is first presented, followed by child information (as reported by the caregivers). The descriptive statistics on participants' GAD-7 (anxiety), PHQ-9 (depression), and CQOLCF (quality of life) scores are then presented. The following sections provide information on the descriptive findings.

Descriptive Statistics of Participant Parents and Caregivers

Of the 297 initial participants, 155 participants fit the proposed inclusion criteria; their demographic information is provided in Table 8. A primary goal of this study was to increase father/male caregiver participation; although I was hoping for a larger turnout of male participants, I was pleasantly surprised with 13 fathers/male caregivers, representing 8.4% of the sample, with the other 91.6% (n = 142) being mothers/female caregivers. A majority of participants were aged 49 or younger (n = 132, 85%), with the 30–39 age group being the most represented (n = 56, 36.4%) and the 60–69 age group being the least represented (n = 4, 2.6%).

Table 8

Parent Caregiver Descriptive Statistic Data (N = 155)

Variable	Frequency (*n*)	Percentage (*%*)
Gender		
Female	142	91.6
Male	13	8.4
Age Group		
21–29	27	17.5
30–39	56	36.4
40–49	49	31.8
50–59	18	11.7
60–69	4	2.6
Race/Ethnicity		
White	142	91.6
Mixed Race	6	3.9
Asian	3	1.9
Black	2	1.3
Hispanic	2	1.3
Relationship Status		
Married/With a Partner	121	78.1
Separated/Divorced	20	12.9
Single	12	7.7
Widowed	2	1.3
Religious Beliefs		
Christian	76	49.0
Atheist/Agnostic	47	30.3
Catholic	17	11.0
Other (e.g., Jewish, Hindu)	15	9.7
Level of Education		
< High School	5	3.2
High School Diploma/GED	14	9.0
Some College (No Degree)	41	26.5
Trade School Certification	8	5.2
Associate's Degree	15	9.7
Bachelor's Degree	39	25.2
Master' Degree	23	14.8
Professional (MD, JD)/ or Doctoral (PhD, EdD) Degree	10	6.4
Employment Status		
Work Full-Time	62	40.0
Work Part-Tame	30	19.4

Stay-at-Home Caretaker	49	31.6
Unemployed	14	9.0
Health Insurance		
Employer-Based	74	47.7
Public Assistance	50	32.3
Other (e.g., private)	31	20.0
Annual Income		
< $25,000	22	14.2
$26,000–$40,000	21	13.5
$40,001–$60,000	25	16.1
$60,001–$80,000	19	12.3
$80,001–$100,000	16	10.3
$100,001–$150,000	22	14.2
Above $150,001	19	12.3
Round-Trip Travel to CF Clinic		
< = 40 miles	43	36.4
41–90 miles	35	29.7
> = 91 miles	40	33.9

As CF is the number one life-shortening, genetic, chronic illness (CFF, 2014b) affecting Caucasians, it was not surprising to see 91.6% (n = 142) of participants identified as White, with the other 8.4% identifying as mixed race (n = 6, 3.9%), Asian (n = 3, 1.9%), Black (n = 2, 1.3%), and Hispanic (n = 2, 1.3%). Over 78% of participants were married or with a partner (n = 121, 78.1%), with the other 21.9% being separated/divorced (n = 20, 12.9%), single (n = 12, 7.7%), and widowed (n = 2, 1.3%). As the participants were predominantly Caucasian, the top three religious belief affiliations were Christian (n = 76, 49%), Atheist/Agnostic (n = 47, 30.3%), and Catholic (n = 17, 11%), with 9.7% (n = 15) identifying as "Other" including Adventist, Hindu, Islam, Jewish, Quaker, and unsure.

Educationally, there was a wide range with "Some College (No Degree)" being the second largest group at 26.5% (n = 41); college-level education had the highest

number of participants with 72 (46%), with some having a bachelor's degree (n = 39, 25.2%), a higher level master's degree (n = 23, 14.8%), or professional/doctoral degree (n = 10, 6.4%). Five participants (3.2%) had less than a high school education, while 14 (9.0%) had a high school diploma/GED, and 23 participants completed a trade school certification or associate degree (n = 8, 5.2%; n = 15, 9.7%, respectively).

A total of 144 participants responded to the annual income question. With level of education being so varied, the reported annual income also had a wide range from less than $25,000 per year ($n$ = 22, 14.2%) to over $150,001 per year ($n$ = 19, 12.3%). There was a wide range in reported gross annual income of participants: from $25,001 to $40,000 annually ($n$ = 21, 13.5%); $40,001 to $60,000 annually ($n$ = 25, 16.1%); $60,001 to $80,000 annually ($n$ = 19, 12.3%); $80,001 to $100,00 annually ($n$ = 16, 10.3%) earned; and $100,000 to $150,000 annually ($n$ = 22, 14.2%). Regarding employment status, participants reported having full-time (n = 62, 40%) versus part-time (n = 30, 19.4%) employment, unemployment (n = 14, 9.0%), and identified themselves as stay-at-home caretakers (n = 49, 31.6%). Based on the employment data, it is not surprising to see that almost half of the participants (n = 74, 47.7%) had employer-based health insurance, with the remaining participants divided nearly equally between private insurance (n = 31, 20%) and public assistance (n = 50, 32.3%).

Although 155 participants were included in the descriptive statistics, only 118 answered the question regarding distance traveled roundtrip for their CF clinic visits. The results for travelling roundtrip fell into three groups: distance being less than or equal to

40 miles (n = 43, 36.4%), distance of 41–90 miles (n = 35, 29.7%), and distance being greater than or equal to 91 miles (n = 40, 33.9%) to their CF clinic.

Participant Experiences with Anxiety, Depression, and other Mental Health History

Next, participants completed questions concerning their mental health and their current and former experiences with anxiety, depression, other diagnosis, therapeutic treatment (counseling/therapy), and use of psychotropic medication. Participant responses varied in number per question, from 148 to 155, and the results of these findings are below and are also presented in Table 9.

Table 9

Parent Reports of Mental Health History

Variable	Frequency (*n*)	Percentage (*%*)
Current Anxiety Diagnosis (*n* = 149)		
No	82	55.0
Yes	67	45.0
Current Depression Diagnosis (*n* = 148)		
No	106	71.6
Yes	42	28.4
Current "Other" MH Diagnosis (*n* = 153)		
No	143	93.5
Yes	10	6.7
Current MH Treatment (*n* = 148)		
No	120	81.1
Yes	28	18.9
Current Psychotropic Medication (*n* = 149)		
No	114	76.5
Yes	35	23.5
Former Anxiety Diagnosis (*n* = 152)		
No	67	44.1
Yes	85	55.9
Former Depression Diagnosis (*n* = 150)		
No	78	48.0
Yes	72	52.0
Former "Other" MH Diagnosis (*n* = 155)		
No	132	85.2
Yes	23	14.8
Former MH Treatment (*n* = 153)		
No	64	41.8
Yes	89	58.2
Former Psychotropic Medication (*n* = 149)		
No	83	55.7
Yes	66	44.3
Interested in MH Check at each Clinic Visit (*n* = 152)		
No	26	17.1
Yes	86	56.6
Unsure	31	20.4
Other	9	5.9

Regarding anxiety, participants reported having a former diagnosis (*n* = 85, 55.9%), and/or as being currently diagnosed (*n* = 67, 45.0%). Regarding depression, participants stated they had a former diagnosis (*n* = 72, 52.0%) and/or as being currently diagnosed (*n* = 42, 28.4%). Almost 15% (*n* = 23) reported they had another former mental health diagnosis (*n* = 23), while 6.7% (*n* = 10) had a current "other" diagnosis such as postpartum depression (PPD), attention deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), posttraumatic stress disorder (PTSD), or personality disorder. Participants reported they had previously received mental health services (*n* = 89, 58.2%), and/or were currently working with a counselor or therapist (*n* = 28, 18.9%). Almost 45% (*n* = 66) reported previous use of psychotropic medication, with only 23.5% (*n* = 35) currently utilizing medication to support their mental health needs.

Parents were also asked about their interest in receiving a mental health checkup during their child's routine CF clinic visits, approximately four times per year, results were as follows: yes (*n* = 86, 56%); no (*n* = 26, 17.1%); unsure (*n* = 31, 20.4%); and "other" (*n* = 9, 5.9%) and provided detailed concerns. Of the nine, three participants reported they would prefer a caregiver mental health screening either annually or biannually. Other responses discussed concern on how the information would be used along with hesitation of being approached – stating this should be a service only if the caregiver "asks for help." On the other hand, two participants shared similar sentiments requesting handouts with information and resources to help caregivers, otherwise they would not be interested in a mental health screening. One participant reported their CF

center already provides this service, but the frequency of the screenings was unclear and left unstated by the participant.

Descriptive Statistics of Caregiver Experiences of Anxiety, Depression, and QOL

Many participants chose not to answer one or more of the PHQ-9, GAD-7, and CQOLCF items. As such, only 100 participants had a CQOLCF composite score, and just 88 participants had PHQ-9 and GAD-7 composite scores. A decision was made to impute the 12 missing data points, from the means of the original 88 participants for the PHQ-9 (M = 10.74, SD = 6.65) and GAD-7 (M = 11.07, SD = 5.93) composite scores using mean imputation. As a result, 100 participants had complete PHQ-9, GAD-7, and CQOLCF scale data.

The PHQ-9 and GAD-7 both have cutoff scoring points of 5, 10, and 15 indicating mild, moderate, and severe levels of depression and anxiety, with 10 being the recommended cutoff point for further evaluation (Kroenke et al., n.d.). The CQOLCF does not have cutoff points for clinical evaluation recommendations, therefore a decision was made to break this in to two categories, with less than 70 indicating low to average quality of life, and greater than 71 indicating average to high quality of life. The decision to artificially divide the quality of life into two groups was based on the median reported score of participants (Md = 71, SD = 18.03) to highlight the differences between groups. A breakdown of results may be seen in Table 10.

Table 10

Rates of Anxiety and Depression in Caregivers (n = 100)

Assessment scoring	Categories	Total (%)	Mother (%)	Father (%)
PHQ-9				
0–4	None to minimal	19	16	44
5–9	Mild	33	34	22
10–14	Moderate	18	18	22
15–19	Moderately severe	19	21	0
20–27	Severe	11	11	12
GAD-7				
0–5	None to minimal	15	15	12
5–9	Mild	28	26	44
10–14	Moderate	27	28	22
15–21	Severe	30	31	22
CQOLCF				
< = 70	Low to average (< = 70)	50	51	44
> = 71	Average to high (> = 71)	50	49	56

Note. As the *n* = 100, frequency and percentage are the same for the total percentages.

It was hypothesized that female caregivers would experience higher rates of anxiety and depression and lower rates of quality of life when compared to their male counterparts. As a whole, the hypothesis was rejected as there were no significant

differences found between male and female caregivers in rates of anxiety, depression, or quality of life. These results could be due to the fact that the sample was skewed favorably toward female participants, who comprised over 91% of the population. Additionally, post hoc chi-square tests on the PHQ-9, GAD-7, and CQOLCF were not significant, likely a result of the small sample of fathers. However, the female mean score (M = 11.10, SE = 0.70) is higher than the male mean score (M = 7.78, SE = 2.21), placing mothers in the moderate depression group and fathers in the mild depression group. Additionally, quality of life mean scores for male participants (M = 81.89, SE = 5.95) was relatively higher than reported scores for female participants (M = 71.24, SE = 1.88).

When looking at the breakdown of the range of scores for depression, a majority 66% of fathers had no depression to mild depression, compared to 50% of mothers. Although fathers experienced higher rates of moderate (22 %) and severe depression (12%) than mother's reports of moderate (18%) versus severe (11%) depression, there were no male participants experiencing moderately severe depression while 21% of mothers fell into that category. Looking at differences in rates of anxiety, 41% of mothers reported mild or no symptoms compared to 56% of fathers. Mothers reported more moderate levels of anxiety (28%) compared to fathers (22%). Mothers also reported more severe anxiety (31%) compared to fathers (n = 22%). Fifty-one percent of mothers reported low to average QOL compared to 44% of fathers with 56% of fathers reporting average to high QOL compared to 49% of mothers.

Descriptive Statistics of Children with Cystic Fibrosis

Participants were asked to identify a referent child with CF and answer questions for the same child throughout the survey; their demographic information is provided below and may be found with full descriptive information in Table 11. The parent/caregiver sample was $N = 155$, however, almost half of the participants did not provide information on their child, and as such, the sample sizes vary greatly per question.

Table 11

Child Descriptive Statistic Data

Variable	Frequency (*n*)	Percentage (%)
Child Gender		
Male	52	64.2
Female	29	35.8
Child Age Group		
0–6 years	22	27.2
7–13 years	36	44.4
14+ years	23	28.4
Child School		
Public School	55	49.1
Private School	9	8.0
Montessori	1	0.9
Homeschool	13	11.6
Other	34	30.4

Eighty-one ($n = 81$) participants provided information on their child's gender and age group, with a 64.2% majority raising a son ($n = 52$) and 35.8% reporting raising a daughter with CF ($n = 29$). The 7–13-year-old age group for participant referent child had the largest representation at 44.4% ($n = 36$), with the other two age groups being almost equally split; 27.2% ($n = 22$) were in the 0–6-year group with 28.4% ($n = 23$) in

the 14 and older group. For the school information of the referent child, 112 participants reported almost half of their children attended public school (n = 55, 49.1%), with others reporting homeschool (n = 13, 11.6%), private school (n = 9, 8.0%), Montessori school (n = 1, 0.9%), and "Other" (n = 34, 30.4%). Those who responded "Other" stated their children were not of school age and went to daycare or identified alternate formats of schooling which included public charter school, being taught in the home by a public-school teacher, or virtual school, most likely due to the pandemic.

Severity of Cystic Fibrosis among Children

Participants were asked to identify their child's current CF severity, age of the child at diagnosis, type of testing to determine CF diagnosis, as well as medical issues and care surrounding their child's CF; this information is provided below with results presented in Table 12.

Table 12

Child Cystic Fibrosis Disease Specific Descriptive Statistics (n = 118)

Variable	Frequency (*n*)	Percentage (*%*)
Child CF Severity		
Mild	56	47.8
Moderate	43	36.8
Severe	18	15.4
CF Diagnostic Test		
Amniocentesis	6	3.9
Chest radiograph (x-ray)	3	1.9
Chorionic villus sampling (CVS; prenatal screening)	1	0.6
Clinical evaluation at CF care center	8	5.2
Genetic carrier test	33	21.3
Immunoreactive trypsinogen (IRT; newborn screening)	7	4.5
Medical test with PCP	2	1.3
Newborn screening for CF	60	38.7
Pulmonary function test	1	0.6
Sputum test	2	1.3
Stool evaluation	11	7.1
Sweat chloride test	68	43.9
Other	14	9.0
Child CF Mutation		
F508del mutation	56	53.3
F508del mutation and additional mutation(s)	49	46.7
FEV_1/Pulmonary Function Test		
91–100	37	32.5
81–90	20	17.5
71–80	5	4.4
61–70	3	2.6
51–60	4	3.5
31–40	2	1.8
< 20	2	1.8
Unknown	41	36.0

Of the 118 participants who answered the CF diagnostic test question, the primary methods of CF diagnosis were sweat chloride test (*n* = 68, 43.9%) and newborn screening for CF (*n* = 60, 38.7%), followed by genetic carrier test (*n* = 33, 21.3%). Eleven (7.1%) received diagnosis from a stool evaluation with 14 (9.0%) responding "Other;" of these, nine identifying bowel obstruction, and/or their child being born with Meconium Ileus, as how they received their child's CF diagnosis. Eight children (5.2%), received confirmation through a clinical evaluation at a CF care center, seven (4.5%) confirmed via immunoreactive trypsinogen, six (3.9%) via amniocentesis, three (1.9%) via chest radiograph (x-ray), two (1.3%) via sputum test or medical test with their PCP, and one (0.6%) via chorionic villus sampling or pulmonary function test.

CF disease severity reported was mild 36.1% (*n* = 56), moderate 27.7% (*n* = 43), and severe 11.6% (*n* = 18), totaling 117 participant responses for this question. When asked to identify the child's primary CFTR gene mutation, 105 participants responded, as follows: F508del mutation only (*n* = 56, 53.3%), and F508del mutation as well as an additional mutation (*n* = 49, 31.6%). Finally, participants were asked questions regarding their child's FEV_1/PFT, or respiratory volume/ pulmonary function test. Pulmonary function is an important diagnostic tool that provides insight into the clinical indication of health of individuals with CF; less than 40% pulmonary function is categorized as severe, 40% to 69% is moderate, 70% to 89% is mild, and 90% and above is normal (CFF, 2021a). A majority of 62 participants (54%) reported that their children were within normal and mild ranges of pulmonary function with PFT values above 70%. Nine (7.9%) participants reported their children had moderate lung function ranging between 40% to

69%, and two (1.8%) reported their child as having severely low lung function falling into the under- 20% category. Interestingly, 41 participants (36%) reported they did not know their child's most recent pulmonary lung function test results.

Physical Health Concerns of Children with Cystic Fibrosis

Participants answered questions concerning the physical health of their child with CF, including recent hospitalizations, tune ups, bacterial infections, and CF-related illness/disease and/or related complications. Participant responses varied in number for each question and are listed accordingly. The results of these findings are below and are also presented in Table 13.

Table 13

Parent Caregiver Reports of Child's Medical History

Variable	Frequency (*n*)	Percentage (%)
Hospitalization in Past 6 Months (*n* = 120)		
No	98	81.7
Yes	22	18.3
Nasal Surgery in Past 6 Months (*n* = 119)		
No	112	94.1
Yes	7	5.9
Lung Transplant List (*n* = 121)		
No	118	97.5
Yes	3	2.5
Lung Transplant Recipient (*n* = 120)		
No	118	98.3
Yes	2	1.7
CF-Related Diabetes (*n* = 121)		
No	101	83.5
Yes	20	16.5
CF-Related Liver Disease (*n* = 121)		
No	106	87.6
Yes	15	12.4
CF-Related Metabolic Syndrome (*n* = 121)		
No	116	95.9
Yes	5	4.1
Positive *Pseudomonas Aeruginosa* Culture (*n* = 121)		
No	50	41.3
Yes	71	58.7
Positive M*ycobacterium Abscessus* Culture (*n* = 121)		
No	116	95.9
Yes	5	4.1
Positive *Burkholderia Cenocepacia C*ulture (*n* = 121)		
No	117	96.7
Yes	4	3.3

Of the 120 participants who provided a response to their child's hospitalization in the past 6 months for a tune up, 98 participants (81.7%) reported that their child was not hospitalized while 22 (18.3%) stated their child did need a hospitalized tune up in the past

6 months. Regarding their child being on a lung transplant list or receiving a lung transplant, 121 and 120 participants responded, respectively, as only three children (2.5%) were on a lung transplant list and only two (1.7%) were lung transplant recipients. Participants (*n* = 121) provided information on their child's CF related illness, disorders, or complications; they reported their child had CF-related asthma (*n* = 36, 29.8%), CF-related diabetes (*n* = 20, 16.5%), CF-related liver disease (*n* = 15, 12.4%), CF-related metabolic syndrome (*n* = 5, 4.1%), and CF-related bone disease (*n* = 4, 3.3%). Participants also completed questions concerning if their child had ever had a positive bacterial culture, of which 121 provided information; more than half reported a positive *pseudomonas aeruginosa* culture (*n* = 71, 58.7%), m*ycobacterium abscessus* culture (*n* = 5, 4.1%), and *burkholderia cenocepacia* culture (*n* = 4, 3.3%).

Mental Health History of Children with Cystic Fibrosis

Next, participants answered questions concerning the mental health of their child with CF, including experiencing anxiety, depression, other diagnosis, therapeutic treatment (counseling/therapy) outside of their CF clinic, and psychotropic medication. Again, responses varied in number per question from 117 to 121 participants, and the results of these findings are below and are also presented in Table 14.

Table 14

Parents Caregiver Reports of Child Mental Health History

Variable	Frequency (*n*)	Percentage (%)
Child Current Anxiety Diagnosis (*n* = 119)		
No	93	78.2
Yes	26	21.8
Child Current Depression Diagnosis (*n* = 117)		
No	104	88.9
Yes	13	11.1
Child Current "Other" MH Diagnosis (*n* = 121)		
No	105	86.8
Yes	16	13.2
Child Current MH Treatment (*n* = 118)		
No	92	78.0
Yes	26	22.0
Child Current Psychotropic Medication (*n* = 119)		
No	107	89.9
Yes	12	10.1
Child Former Anxiety Diagnosis (*n* = 121)		
No	82	67.8
Yes	39	32.2
Child Former Depression Diagnosis (*n* = 118)		
No	99	83.9
Yes	19	16.1
Child Former "Other" MH Diagnosis (*n* = 121)		
No	110	90.9
Yes	11	9.1
Child Former MH Treatment (*n* = 120)		
No	76	63.3
Yes	44	36.7
Child Former Psychotropic Medication (*n* = 118)		
No	105	89.0
Yes	13	11.0
Interested in MH Check at each Clinic Visit (*n* = 118)		
No	11	9.3
Yes	81	68.6
Unsure	22	18.6
Other	4	3.4

Regarding anxiety, caregivers identified their child as having a former diagnosis (*n* = 39, 32.2%) and/or as being currently diagnosed (*n* = 26, 21.8%). Regarding depression, caregivers identified their child as having a former diagnosis (*n* = 19, 16.1%) and/or as being currently diagnosed (*n* = 13, 11.1%). A little over 9% (*n* = 11) reported their child had another former mental health diagnosis, while 13.2% (*n* = 16) had a current "other" diagnosis such as ADD, autism, OCD, PTSD, and bipolar depression. Participants reported their children as previously receiving mental health services (*n* = 44, 36.7%), as well as currently receiving mental health treatment outside of their CF care center with (*n* = 26, 22%). Participants reported their child was previously prescribed psychotropic medications to treat mental health conditions (*n* = 13, 11.0%), and/or as currently utilizing psychotropic medications to treat mental health conditions (*n* = 12, 10.1%).

Parents were also asked about their interest in their child receiving a mental health checkup at their routine CF clinic visits, approximately four times per year. A majority said yes (*n* = 81, 68.6%), some said no (*n* = 11, 9.3%), a good amount were unsure (*n* = 22, 18.6%), and a small percentage (*n* = 4, 3.4%) replied "other" and provided detailed concerns. Participants stated they would like this service for their children as well as themselves and some stated they would like a biannual checkup opportunity. Other participants reported an age limited of 10 years old as when they would like mental wellness checks to begin, while one participant stated it would "depend on if there are resources my child could benefit from."

Dependent Variable Descriptive Statistics

The quantitative component of this study focused on three parent/caregiver dependent variables including: (a) participant anxiety, measured by the GAD-7; (b) participant depression, assessed by the PHQ-9; and (c) participant quality of life, measured by the CQOLCF. The data results showed a total of 100 participants as completing the GAD-7, PHQ-9, and CQOLCF scales; for reporting consistency purposes, the findings are presented for these 100 participants in Table 15.

Table 15

Descriptive Statistics: Dependent Variables (N = 100)

Assessment measure	*M*	*Md*	*SD*	*Min*	*Max*	$Z_{skewness}$	Cronbach's α
GAD-7: Anxiety	11.07	10.50	5.93	0	21	0.35	.93
PHQ-9: Depression	10.74	9.00	6.65	0	27	2.59	.89
CQOLCF: CF Quality of Life	72.21	71.00	18.03	28	109	-0.58	.90

Note. $z_{skewness}$ = skewness/SE skewness. Should be < 3.29 (Kim, 2013).

The participants' mean score on the GAD-7 was 11.07 (*Md* = 10.50, *SD* = 5.93), equivalent to indications of moderate levels of anxiety. The participants' PHQ-9 mean score was 10.74 (*Md* = 9.00, *SD* = 6.65), equivalent to indications of moderate levels of depression. The participants' CQOLCF mean score was 72.21 (*Md* = 71.00, *SD* = 18.03), indicating low-to-moderate levels of quality of life.

The $z_{skewness}$ values (i.e.., skewness/SE skewness; Kim, 2013) were computed to determine if the assumption of variable normality was met for the three variables. The assumption of normality is met if the $z_{skewness}$ value is less than +/-3.29 (Kim, 2013). As

noted in Table 14, the GAD-7 had a $z_{skewness}$ value of 0.35, the PHQ-9 had a $z_{skewness}$ value of 2.59 and the CQOLCF quality of life scale had a $z_{skewness}$ value of -0.58, denoting that the normality assumption was met for all three variables. Cronbach's alphas (α) were also computed to determine the inter-item reliability of the variables. The GAD-7 anxiety scale had a Cronbach's α of .93, indicating excellent inter-item reliability. The PHQ-9 had a Cronbach's α of .89 and the CQOLCF quality of life scale had a Cronbach's α of .88, denoting very good reliability.

Hypothesis Testing

A series of one-way and factorial MANOVAs were conducted to answer the quantitative research questions. The hypothesis testing involved analyzing data from the 100 participants who completed the GAD-7, PHQ-9, and CQOLCF scales. This section of the chapter provides the findings from the MANOVA analyses.

Research Question One

The first research question was "How do female caregivers and male caregivers raising a child with a cystic fibrosis diagnosis differ in rates of anxiety, depression, and quality of life?" The H_a1 states: Female caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression), and lower scores of CQOLCF (quality of life) when compared to scores of male caregivers. A one-way MANOVA was conducted to address this question by determining if there were significant participant gender differences concerning caregiver anxiety, depression, and quality of life.

Results from the MANOVA are presented in Table 16. The overall model was not significant, *Wilks* λ = .964, $F(3, 95) = 1.19$, $p = .318$. There were no significant gender differences on participants' GAD-7 anxiety scores, $F(1, 98) = 0.68$, $p = .413$, PHQ-9 depression scores, $F(1, 98) = 2.06$, $p = .155$, or CQOLCF quality of life scores, $F(1, 98) = 2.91$, $p = .091$. Although there were no significant gender differences regarding participant reported scores of emotional health, female participant mean scores of anxiety and depression reflect moderate levels of both indicating further clinic intervention as recommended. Female mean scores of anxiety ($M = 11.22$, $SE = 0.63$) were higher than male participants ($M = 9.56$, $SE = 1.98$). Mean scores of depression were higher for mothers ($M = 11.10$, $SE = 0.70$) than fathers ($M = 7.78$, $SE = 2.21$). Female participants also had a lower quality of life mean scores ($M = 71.24$, $SE = 1.88$) when compared to male participants ($M = 81.89$, $SE = 5.95$).

Table 16

One-way MANOVA: Parent Gender and Anxiety, Depression, and CF Quality of Life (n = 100)

Dependent variable	*Df*	*F*	*p*	Gender	*M*	*SE*	95% CI	
							Lower	*Upper*
Anxiety	1,98	0.68	.413	Male	9.56	1.98	5.63	13.48
				Female	11.22	0.63	9.98	12.46
Depression	1,98	2.06	.155	Male	7.78	2.21	3.40	12.16
				Female	11.10	0.70	9.66	12.41
CF Quality of Life	1,98	2.91	.091	Male	81.89	5.95	70.08	93.70
				Female	71.24	1.88	67.51	74.98

Post hoc chi-square testing was conducted to further assess if there were significant participant gender differences concerning caregiver anxiety, depression, and quality of life. There were no significant differences between mothers and fathers for PHQ-9 (depression), $\chi^2(4) = 5.70$, $p = .223$, GAD-7 (anxiety), $\chi^2(4) = 2.42$, $p = .658$, or (quality of life), $\chi^2(1) = 0.12$, $p = .727$. Due to the overall lack of significant findings, the H_a1 that female caregivers will have higher scores on the GAD-7 (anxiety), PHQ-9 (depression), and lower scores on the CQOLCF (quality of life) when compared to scores of male caregivers was rejected.

Research Question Two

The second research question was, "How does a referent child's CFTR genetic mutation and CF severity relate to caregiver rates of anxiety, depression, and quality of life?" The H_a2 states: Caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression) and lowers scores on the CQOLCF (quality of life) when compared to their child's CFTR genetic mutation and CF severity; caregivers of children with greater CF severity and rarer genetic mutations will have higher scores. To address the second research question, a 2 (CF mutation type) by 3 (CF severity) factorial MANOVA was conducted to determine if there were significant child CF mutation and child CF severity effects on parents' anxiety, depression, and quality of life. As the analysis was a factorial MANOVA, results provided information on the main effects of child gender and age group and the interaction effects of child gender by age group.

The MANOVA results findings for the main effects of child CF mutation type and child CF severity category are presented in Table 17. The overall model for child CF genetic mutation was not significant, *Wilks* $\lambda = .986$, $F(3, 77) = 0.38$, $p = .771$. There were no significant child CF genetic mutation categorical differences concerning parents' GAD-7 anxiety scores, $F(1, 79) = 0.07$, $p = .786$, PHQ-9 depression scores, $F(2, 79) = 0.39$, $p = .536$, and CQOLCF quality of life scores, $F(1, 79) = 1.02$, $p = .315$.

Table 17

Factorial MANOVA: Child CF Genetic Mutation and CF Severity and Parents' Depression, Anxiety, and Quality of Life (N = 100)

Dependent variable	*Df*	*F*	*p*	IV	*M*	*SE*	95% CI	
							Lower	*Upper*
Anxiety	1,79	0.07	.786	F508del Only	11.01	0.94	9.14	12.88
				F508del plus addtl CFTR	10.60	1.18	8.24	12.89
	2,79	0.91	.407	Mild CF	10.79	0.89	9.03	12.56
				Moderate CF	12.11	1.05	10.02	14.20
				Severe CF	9.52	1.80	5.94	13.10
Depression	1,79	0.39	.536	F508del Only	10.94	1.06	8.84	13.04
				F508del plus addtl CFTR	9.82	1.32	7.20	12.44
	2,79	1.22	.301	Mild CF	11.48	0.99	9.50	13.46
				Moderate CF	11.60	1.18	9.26	13.94
				Severe CF	8.16	2.02	4.14	12.19
CF Quality of Life	1,79	1.02	.315	F508del Only	70.96	2.82	65.34	76.58
				F508del plus addtl CFTR	75.53	3.53	68.51	82.54
	2,79	2.83	.065	Mild CF	73.41	2.65	68.12	78.69
				Moderate CF	66.36	3.14	60.11	72.60
				Severe CF	79.96	5.39	69.25	90.68

The effects of child CF severity category on participants' anxiety, depression, and quality of life are presented in Table 17. The overall MANOVA model for child CF

severity category was not significant, *Wilks* λ = .897, *F(*6, 156) = 1.44, p = .203. There were no significant child CF severity categorical differences concerning parents' GAD-7 anxiety scores, $F(2, 79) = 0.71$, p = .495, PHQ-9 depression scores, $F(2, 79) = 1.21$, p = .301, or CQOLCF quality of life scores, $F(2, 79) = 2.83$, p = .065.

The child CF genetic mutation by child CF severity interaction effects were not significant. Factorial MANOVA findings showed no significant child CF genetic mutation by child CF severity category interaction effects on parents' GAD-7 scores, $F(2, 79) = 0.71$, p = .495, PHQ-9 depression scores, $F(2, 79) = 0.53$, p = .592, or CQOLCF quality of life scores, $F(2, 79) = 0.50$, p = .608.

Although no statistical significance was found, it is important to note that anxiety and depression levels reported were similar for genetic mutation F508del Only (M = 11.01, SE = 094; M = 10.94, SE = 1.06, respectively) compared to F508del plus additional CFTR mutation (M = 10.60; SE = 1.18; M = 9.82, SE = 1.32, respectively) with very little difference reported. Additionally, the child's genetic mutation also appeared to have little impact on parental quality of life with F508del only (M = 70.96, SE = 2.82) and F508del plus additional mutations (M = 75.53, SE = 3.53) having minimal numerical differences in range. Reported disease severity also had very little impact on caregiver emotional health. Caregiver reported rates of anxiety were similar for mild (M = *10*.79, SE = 0.89), moderate (M = 12.11, SE = 1.05), and severe (M = 9.52, SE = 1.80) disease severity. Reported rates of caregiver depression were also similar to that of anxiety for mild (M = 11.48, SE = 0.99), moderate (M = 11.60, SE = 1.18), and severe (M = 8.16, SE = 2.02) disease severity. Finally, quality of life had a larger range compared to rates of

anxiety and depression, although the reported rate differences were still relatively small for mild (M = 73.41, SE = 2.65), moderate (M = 66.36, SE = 3.14), and severe (M = 79.69, SE = 5.39) disease severity.

The alternative hypothesis, H_a2, that caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression), and lower scores on the CQOLCF (quality of life) when compared to their child's CFTR genetic mutation and CF severity and caregivers of children with greater CF severity and rarer genetic mutations will have higher scores was rejected based on the non-significant findings.

Research Question Three

The third research question was, "How does the referent child's age and sex affect caregiver anxiety, depression, and quality of life?" The H_a3: states: Caregivers with infants and female adolescent children will have higher scores on the GAD-7 (anxiety), PHQ-9 (depression) and lower scores on the CQOLCF (quality of life). To address the third research question, a 2 (child gender) by 3 (child age group) factorial MANOVA was conducted to determine if there were significant child gender and age group differences concerning anxiety, depression, and quality of life in the sample of participants of children with CF. As the analysis was a factorial MANOVA, results provided information on the main effects of child gender and age group and the interaction effects of child gender by age group. As main and interaction effects were found to be significant, there are two separate MANOVA tables, one for main effects of child gender and age group (see Table 18) and one for the interaction effects of child gender by age group (see Table 19).

Table 18

Factorial MANOVA: Child Gender and Age Group and Parents' Anxiety, Depression, and Quality of Life (N = 100)

Dependent variable	*Df*	*F*	*p*	IV	*M*	SE	95% CI	
							Lower	*Upper*
Anxiety	*1,75*	*4.20*	*.044*	Male	11.77	0.75	10.27	13.27
				Female	9.02	1.11	6.80	11.23
	2,75	*6.53*	*.002*	0–6 years	13.44	1.19	11.07	15.81
				7–13 years	10.81	0.90	9.02	12.61
				14+ years	6.93	1.36	4.23	9.63
Depression	1,75	2.05	.156	Male	11.24	0.89	9.46	13.02
				Female	8.96	1.32	6.33	11.59
	2,75	*3.86*	*.025*	0–6 years	12.81	1.41	10.00	15.62
				7–13 years	10.60	1.07	8.47	12.73
				14+ years	6.89	1.61	3.68	10.10
CF Quality of Life	1,75	3.03	.086	Male	69.07	2.53	64.04	74.11
				Female	76.93	3.74	69.48	84.38
	2,75	2.91	.061	0–6 years	65.58	3.99	57.62	73.58
				7–13 years	73.39	3.02	67.37	79.41
				14+ years	80.04	4.56	70.96	89.12

Note. Significant findings in italics.

The MANOVA results findings for the main effects of child gender and age group are presented in Table 18. The overall model for child gender was not significant, *Wilks λ*

= .932, $F(3, 73) = 1.77$, $p = .160$. However, there were significant child gender differences concerning parents' GAD-7 anxiety scores, $F(1, 75) = 4.20$ $p = .044$. Caregivers of male children reported significantly higher levels of anxiety ($M = 11.77$, $SE = 0.75$) when compared to caregivers of female children ($M = 9.02$, $SE = 1.11$). There were no significant child gender effects concerning participants' PHQ-9 depression scores, $F(1, 75) = 2.05$, $p = .156$, or parents' CQOLCF quality of life scores, $F(1, 75) = 3.03$, $p = .086$.

The overall MANOVA model for child age group was significant, *Wilks* $\lambda = .836$, $F(6, 146) = 2.28$, $p = .039$. There were significant child age group differences concerning participants' GAD-7 anxiety scores, $F(2, 75) = 6.53$, $p = .002$. Tukey *post hoc* tests showed that parents of children ages 0–6 years old reported significantly higher levels of anxiety ($M = 13.44$, $SE = 1.19$) when compared to parents of children ages 7–13 years old ($M = 10.81$, $SE = 0.90$) and parents of children 14 or older ($M = 6.93$, $SE = 1.36$). There were significant child age group differences concerning participants' PHQ-9 depression scores, $F(2, 75) = 3.86$, $p = .025$. Tukey *post hoc* tests showed that caregivers of children ages 0–6 years old reported significantly higher levels of depression ($M = 12.81$, $SE = 1.41$) as compared to caregivers of children ages 7–13 years old ($M = 10.60$, $SE = 1.07$) and caregivers of children 14 or older ($M = 6.89$, $SE = 1.61$). There was not a significant difference concerning child age group differences and parents' CQOLCF quality of life scores, $F(2, 75) = 2.91$, $p = .061$.

Table 19

Factorial MANOVA: Child Gender by Age Group Interaction and Parents' Anxiety, Depression, and Quality of Life (n = 100)

Dependent variable	*Df*	*F*	*p*	Gender	Age group	*M*	*SE*	95% CI *Lower*	95% CI *Upper*
Anxiety	*2,75*	*3.65*	*.031*	Male	0–6	13.50	1.43	10.64	16.36
					7–13	10.75	1.20	8.36	13.14
					14+	11.06	1.27	8.54	13.58
				Female	0–6	13.38	1.90	9.60	17.15
					7–13	10.88	1.34	8.20	13.55
					14+	2.80	2.40	1.98	7.58
Depression	2,75	2.58	.082	Male	0–6	12.00	1.70	8.61	15.39
					7–13	10.95	1.42	8.11	13.79
					14+	10.78	1.50	7.79	13.77
				Female	0–6	13.63	2.25	9.14	18.11
					7–13	10.25	1.59	7.08	13.42
					14+	3.00	2.85	2.67	8.68
CF Quality of Life	2,75	2.89	.062	Male	0–6	62.29	4.82	52.69	71.88
					7–13	75.65	4.03	67.62	83.68
					14+	69.28	4.25	60.81	77.74
				Female	0–6	68.88	6.37	56.18	81.57
					7–13	71.13	4.51	62.15	80.10
					14+	90.80	8.06	74.74	106.86

Note. Significant findings in italics.

The factorial MANOVA also provided results for the child gender by age groups, which is presented in Table 19. The overall MANOVA model for the child gender by age group interaction effect was not significant, *Wilks* $\lambda = .856$, $F(6, 144) = 1.97$, $p = .074$. However, univariate findings showed a significant child gender by age group effect on participants' GAD-7 anxiety scores, $F(2, 75) = 3.65$, $p = .031$. Caregivers of girls ages 14 or older reported the lowest levels of anxiety ($M = 2.80$, $SE = 2.40$), while caregivers of boys and girls ages 0–6 years old reported significantly higher levels of anxiety ($M = 13.50$, $SE = 1.43$; $M = 13.38$, $SE = 1.90$, respectively) as compared to caregivers of boys 14 or older ($M = 11.06$, $SE = 1.27$), caregivers of girls 7–13 years old ($M = 10.88$, $SE = 1.34$), and caregivers of boys ages 7–13 years old ($M = 10.75$, $SE = 1.20$).

No significant child gender by age group differences were found concerning participants' PHQ-9 depression scores, $F(2, 75) = 2.58$, $p = .082$. Although there were no significant findings, it is important to note, the interaction observed for caregivers raising daughters ages 14 or older reported significantly lower rates of depression ($M = 3.00$, $SE = 2.85$) compared to all other age groups. Parents raising daughters ages 0–6 years old reported the highest levels of depression ($M = 13.63$, $SE = 2.25$) as compared to caregivers of girls 7–13 years old ($M = 10.25$, $SE = 1.59$), caregivers of boys 14 or older ($M = 10.78$, $SE = 1.50$), and caregivers of boys ages 7–13 years old ($M = 10.95$, $SE = 1.42$), and caregivers of boys ages 0–6 years old ($M = 12.00$, $SE = 1.70$).

No significant child gender by age group differences were found concerning participants' participants' CQOLCF quality of life scores, $F(2, 75) = 2.89$, $p = .062$. With the lack of significance, it is still important to note that caregivers of daughters 14 and

older reported the highest level of quality of life (M = 90.80, SE = 8.06) and caregivers of boys ages 0–6 years old reported the lowest levels of quality of life (M = 62.29, SE = 4.82). Mean reported scores of quality of life were similar across other age groups for caregivers raising sons ages 7–13 years old (M = 75.65, SE = 4.03), sons 14 and older (M = 69.28, SE = 4.25), and caregivers raising daughters ages 0–6 years old (M = 68.88, SE = 6.37) and daughters ages 7–13 years old (M = 71.13, SE = 4.51). Due to the nonsignificant findings concerning the main effects of child gender and age group, the alternative hypothesis, H_a3: Caregivers with infants will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression) and lower scores on quality of life (CQOLCF), was rejected. Additionally, the alternative hypothesis argument that caregivers of female adolescents would have higher PHQ-9 and GAD-7 scores, and lower CQOLCF scores, was not retained.

Research Question Four

The fourth research question was, "How does access to CF clinics affect caregiver anxiety, depression, and quality of life?" The H_a4 stated: Scores of caregivers on the GAD-7 (anxiety), PHQ-9 (depression), and CQOLCF (quality of life) will be negatively impacted by distance from CF clinic. A one-way MANOVA was conducted to determine if there were significant CF clinic distance differences concerning caregivers' anxiety, depression, and quality of life in the sample of 100 participants.

Results from the one-way MANOVA are discussed and presented in Table 20. The overall model was not significant, *Wilks* λ = .935, $F(6, 188) = 1.06$, p = .386. Findings showed that there were no significant CF clinic distance categorical differences on

participant's GAD-7 anxiety scores, $F(2, 95) = 2.11$, $p = .127$, PHQ-9 depression scores, $F(2, 95) = 2.36$, $p = .100$, or CQOLCF quality of life scores, $F(2, 95) = 0.35$, $p = .709$. Although the findings are not significant, reported anxiety mean scores for parents commuting 41-90 miles ($M = 13.00$, $SE = 1.11$) were slightly higher than those driving less than 40 miles ($M = 10.21$, $SE = 0.94$) and more than 91 miles ($M = 10.42$, $SE = 1.06$). Additionally, reported depression mean scores for parents commuting 41-90 miles ($M = 12.89$, $SE = 1.24$) were slightly higher than those driving less than 40 miles ($M = 9.41$, $SE = 1.05$) and more than 91 miles ($M = 10.39$, $SE = 1.18$). Reported quality of life mean scores for parents commuting more than 91 miles ($M = 74.13$, $SE = 3.28$) were slightly higher than those driving less than 40 miles ($M = 72.21$, $SE = 2.92$) and those driving 41-90 miles ($M = 70.18$, $SE = 3.45$). Due to the non-significant findings, the alternative hypothesis, H_a4, scores of caregivers on the GAD-7 (anxiety), PHQ-9 (depression) and CQOLCF (quality of life) will be negatively impacted by distance from CF clinic, was rejected. A summary of alternative hypothesis testing results may be found in Table 21.

Table 20

One-way MANOVA: Travel Distance to Clinic and Parents' Anxiety, Depression, and Quality of Life (N = 100)

Dependent variable	*Df*	*F*	*p*	Clinic distance	*M*	*SE*	95% CI	
							Lower	*Upper*
Anxiety	2,95	2.11	.127	< = 40 miles	10.21	0.94	8.33	12.08
				41–90 miles	13.00	1.11	10.79	15.21
				> = 91 miles	10.42	1.06	8.32	12.52
Depression	2,95	2.36	.100	< = 40 miles	9.41	1.05	7.33	11.49
				41–90 miles	12.89	1.24	10.43	15.35
				> = 91 miles	10.39	1.18	8.05	12.72
CF Quality of Life	2,95	0.35	.709	< = 40 miles	72.21	2.92	66.41	78.00
				41–90 miles	70.18	3.45	63.34	77.02
				>= 91 miles	74.13	3.28	67.63	80.63

Table 21

Summary of Alternative Hypothesis Testing

Alternative hypotheses for research questions 1-4	Outcome: retained or rejected
H_a1: Female caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression), and lower scores of CQOLCF (quality of life) when compared to scores of male caregivers.	Rejected (Table 16)
H_a2: Caregivers will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression) and lower scores on the CQOLCF (quality of life) when compared to their child's CFTR genetic mutation and CF severity; caregivers of children with greater CF severity and rarer genetic mutations will have higher scores.	Rejected (Table 17)
H_a3: Caregivers with infants and female adolescent children will have higher scores on the GAD-7 (anxiety) and PHQ-9 (depression) and lower scores on the CQOLCF (quality of life).	Rejected (Table 18 & Table 19) Overall model for child gender was rejected; Univariate findings were significant for child gender on participant GAD-7 scores. (Table 18) Overall model for child age group was significant. There were significant child age group interaction differences concerning participant anxiety and depression scores. (Table 19)
H_a4: Scores of caregivers on the GAD-7 (anxiety), PHQ-9 (depression), and CQOLCF (quality of life) will be negatively impacted by distance from CF clinic.	Rejected (Table 20)

Qualitative Findings

The qualitative questions responses were used to explore, identify, and understand the biopsychosocial factors that contributed to the increased rates of anxiety and depression and lower levels of quality of life for caregivers within the CF community. The five experiential questions, within the survey, focused on specific topics, including barriers to seeking and receiving mental health services, experiences with anxiety and depression, quality of life contributions, and views on CFTR gene modulation medication. The overall goal in coding was to identify key phrases and emergent patterns to determine the narrative categories that comprised each theme. Throughout the coding process, five experiential themes emerged that were present from participant responses including: (1) The CF Priority, (2) Resources and Support, (3) Pandemic Amplification of Distress, (4) Natural Skepticism and Cautious Optimism, and (5) Barriers to Seeking and Receiving Mental Health Services. A breakdown of the five main themes, along with their subthemes, may be found in Table 22. Each theme will be presented with accounts from participant caregivers in the pages below.

Table 22

Experiential Themes and Subthemes

Theme	Subtheme
The CF Priority	Daily CF Concerns
	Prevention of Spontaneity
	Balancing CF Care Needs
	Grief and Ambiguous Loss
Resources and Support	Time and Money
	Limited Social and Familial Support
	Overall Family Health and Safety
Pandemic Amplification of Distress	Physical Health Concerns
	Mental Health Concerns
Natural Skepticism & Cautious Optimism	Skepticism & Optimism Toward CFTR Gene Modulating Medications
Barriers to Seeking and Receiving Mental Health Services	No Time or Money, Insurance or Support
	Mental Health Stigma
	Access to Mental Health Care Professionals
	Natural Skepticism Toward Mental Health Care

Participant responses in the qualitative, experiential question section of the study ranged from 82 to 56 responses, from questions 1–5, respectively. In my initial coding process, I removed all identifying information of the participants. Therefore, all caregiver demographic information was blinded as I sorted through the data. It was not until I had identified emergent keywords patterns phrases and themes that I went back through to determine if the response provided were from a female caregiver versus male caregiver perspective. There are more participant responses from female caregivers in this chapter because female respondents outnumbered male respondents 142 to 13. For the sake of consistency and ease for the reader, unless noted, participant responses are from female caregivers, predominantly mothers, based on the high percentage of their representation

within the study. At no point in the online survey were participants asked to provide the names of themselves or their children, however, in the qualitative responses, some participants did include this information. All names included in participant responses have been removed and changed to "daughter" or "son" in order to further protect their privacy. Additionally, all participant responses were primarily left in the format in which they were received from the online survey, with their original grammar, spelling, punctuation, etc., unless noted. This chapter will focus on these findings with participant responses to support the themes.

The CF Priority

The first theme, "The CF Priority," was present from the start as caregiver reports indicated it was always on their mind; survival and "keeping their children alive" were a constant and immediate focus. Four subthemes were identified throughout the coding process including: (1) Daily CF Concerns, (2) Prevention of Spontaneity, (3) Balancing CF Care Needs, and (4) Grief and Ambiguous Loss. The following participant accounts provide evidence to support each of the subthemes within this theme.

Daily CF Care Concerns

Daily preparation and planning of treatments and sanitization needs, nutritional concerns, coordinating doctor's appointments and clinic visits were also generally taking up mental space of the caregiver. For the participants in this study, 119 responded to the question regarding the daily time needed to dedicate to administering medication, planning nutrition, and sanitizing with the average amount of time being 3 hours. One participant reported spending 15 minutes per day while another reported fifteen hours per

day; clearly, the range varies greatly and can potentially be explained by the age and needs of the child. Nearly half of the participants (46%) reported their child's daily treatments generally took two and three hours while 25% reported needing either one or four hours. Numerous participants cited these concerns as a troublesome part of their daily life routine and felt they were an obstacle in seeking their own mental health care. One participant reported simply:

> CF care itself takes a lot of time.

Another stated:

> The hardest part is filling in treatments with a busy life schedule. It's hard to find a balance for activities he can participate in, consistent treatments, and enough sleep.

One discussed:

> Lack of time. Already spend too much time in hospitals and clinics. Too many appointments.

One mother identified struggling to manage finding uninterrupted time to complete treatments:

> I can't just play with my kids without having to stop to do treatments or they want a snack then it's meds and I constantly worry am I leaving out the non-CF child.

Prevention of Spontaneity

Many expressed the inability to be spontaneous due to the physical health needs of their CF diagnosed child were frustrating and overwhelming. One mother participant stated:

> When he was younger it was a major time commitment for me. We could no longer just "pick-up and go," everything required planning. The vast number of medications can be overwhelming and I learned to be an advocate for my child in regards to the medications (interactions and allergies). Now my time is freeing up as he is taking more responsibility. I still have to remind him, but he is taking more control over his treatments.

Another mother responded:

> So much more planning just to daily living and treatments, no longer able to just get up and take a random trip, everything needs to be planned for. Having to limit visitors during the regular cold and flu season, now completely having no visitors because of COVID. Admissions are difficult for child care for other children and making sure they are getting their therapies, appointments, etc. Not getting much time away from the hospital because it all falls on you.

While another expressed frustration of scheduling their lives around CF:

> My kids are young and hardly exhibit any symptoms and I fear the unknown so much. We can't leave the house without meds, any overnights need a whole suitcase of equipment and medicine. I feel like we plan our life around their disease. Germs, bacteria. Have to be so cautious.

One mother discussed how the inability to be spontaneous was now an expected part of their lives:

> It's just extra steps to go camping or go to others house because she has all her medications and medical equipment. She went to summer camp and it took an

> extra hour to get her signed in because I had to fill out separate forms for each of her 10 medications. We all have to be conscious of not putting her at risk of getting sick or further her disease which puts strain on family.

Balancing CF Care Needs

Participants struggled not only with life balance, but also the idea of keeping their children in a "CF bubble" verses letting them live a "normal" life. One participant discussed the life balance struggle in a succinct manner:

> It's hard to find the 'life balance'. Trying to manage all of the demands from so many directions.

One mother provided a transitional perspective to her ideas on the bubble as her daughter began Trikafta:

> I have anxiety around work. Around my current home life situation. I also have anxiety around my children's well being and health, especially with my daughter who has CF. I started relaxing a bit with my anxiety around this once she started taking Trikafta, but then shortly after that COVID-19 hit and has raised the anxiety for her again. I worry that I am not doing enough to keep her away from catching anything, and then I worry that I am keeping her too much in a bubble. It is a fine balance and one that is a struggle to navigate because it us ultimately up to me, and if anything were to happen to her I would feel ultimately responsible and at fault.

Another mother highlighted her struggle with balance, the CF bubble, the pandemic, and raising multiple children with atypical needs:

> I had a friend diagnosed with COVID19 only days after coming to our house. Our family tested negative (thank God), but the paranoia and anxiety surrounding the unknowns with our results... and the guilt and self-directed anger for letting someone in our 'bubble' ate at me for days. I'm still pretty unsure how to proceed, as my adult stepkids come over multiple times per week, and each of them are out in the workforce. I can't convince my husband to not have them over (totally understandable). But I want to mega-isolate now... back the way we did in March when this all entered our state. I have to teach my kids from home (both our choice, and the school system is only open for virtual learning) which adds a ton of stress, as my kids are resistant! My 2 older boys both have aspergers and are near genius level, so it's a real trip trying to work on school at home with them. My CF'r is “normal" and needs more help than the other 2. The unknowns about COVID19 are eating me alive.

Grief and Ambiguous Loss

Finally, the ambiguous loss of the life participants had imagined for themselves, and the anticipatory grief they faced due to the uncertainty of how their child would fare in the world that day, or if their disease and lung function would drastically decline unexpectedly, weighed heavily on the hearts of these devoted and dedicated parents. In discussing how their child’s CF diagnosis has changed their life past, present, and future, one mother provided an emotionally vulnerable response touching not only the daily changes but the ambiguous loss she experiences as well:

> I wasn't able to take him home for three months. We live in Idaho; he was life flighted to Salt Lake City, Utah. It was traumatic. He could only eat a tiny bit of milk; I wasn't able to breastfeed. Only pump, it was the worst point of my entire life. He had to have two surgeries. Even as I am answering this, I am emotional... I believe in God, that is the only thing that got me through and keeps me going. All of my dreams have changed but I have hope with trikafta. I will do anything it takes to keep my son alive.

A few participants discussed either their grief or anticipatory grief as directly contributing to their feelings of depression, providing responses such as:

> Finances, my marriage and my grief.

One heartbreaking response:

> Complicated grief from losing my 13 year old son who had CF. Anxious about losing another child, COVID-19, schooling and trying to fit everything into the day while still giving my children time to just be kids. Finances and my marriage.

Another participant shared their struggles:

> A close family friend committed suicide last year and I'm still overcome with grief, just depressed with the isolation and changes from pandemic.

One participant discussed her struggle with her own grief, mental health, her barriers to treatment, and proposed a solution:

> We need grief groups for the living... not just those who've died. I have searched and searched for an appropriate group, and the closest I come is the Facebook CF moms groups I've found. Otherwise, I see a therapist regularly, and we probably

> talk about CF 50% of the time. I was hospitalized once in 2015 at St Vincent Stress Center - I had a 4 day stay and 11 weeks of Intensive outpatient therapy. I was having angel of mercy ideations about my son who was vomiting 3-4x a day (once always between 2-4am.) Of course suicidal ideations would have proceeded anything I may have done to him. Thankfully, the social worker at the children's hospital was able to listen to my scary thinking, and suggested self admission. I was finally able to get a respite nurse to come in through the medicaid waiver, and she helps with all the tedious work, which has helped a lot. She is the reason I only have about 2 hours of work per day for my little man. I am finally taking care of myself now too.

One mother disclosed:

> I think I was depressed for years when my son was young. My doctor called it 'anticipatory grief.' For years I had nightmares about forgetting or losing my son in a variety of situations. Thankfully through talking about my fears of losing my son to Cystic Fibrosis they went away. I started to appreciate my time with him and not focus on what the future may bring. Like I said before, I had 10 years of excellent therapy lol.

Her response is also an insight into the cautious optimism many of the participants expressed throughout their responses, which will be discussed later in the findings. Finally, one mother's response highlighted almost all the obstacles listed above when asked about her contributing factors to anxiety:

> Not enough time in the day for treatments, school, dinner. How am I going to do everything? What if they get more sick? What if they need so much care and hospitalizations? What if they got on Trikafta and it kills their liver? What if they die?

Resources and Support

From the first theme of the CF Priority, it became apparent that this group of individuals also struggled with a lack of resources as well as social and familial support; thus, the second theme, "Resources and Support," will be discussed as our next theme. The subthemes identified within this theme are (1) Time and Money, (2) Limited Social and Familial Support, and (3) Overall Family Health and Safety. While as a blanket statement, this may appear to be representative of many American families, one blinding difference adding to the lack of resources available to this community may be seen in the amount of time and money spent on routine treatments and medical care. As mentioned in The CF Priority, many parents reported struggling with life balance. While this may be a struggle for anyone in the population, parents of children with cystic fibrosis are faced with substantial additional stressors. These include the need for financial resources to cover their child's very costly monthly treatments and medications, finding time to incorporate daily treatments and medical care (up to four hours each day), and balancing the desire to receive social/familial support with the fears of germ exposure and contamination influenced their perceptions of support and resources available.

Time and Money

Lack of time and money were the two most largely discussed concerns regarding resources and support. One father simply stated his biggest obstacles as:

> Lack of time and finances.

One mother reported this was the top concern contributing to her current feelings of anxiety:

> Children. Money getting right drugs for my kids.

Another mother reported time struggles with treatments and how it was affecting her own health:

> Just fitting quality, daily cf care into a schedule with work and distance learning has given me raging insomnia. I haven't slept more than 2 hours at a time in the last two weeks.

The time and financial stressors of cystic fibrosis in general were the most commonly mentioned contributing factors to a majority of the study's participants and were also brought up by many as top obstacles to seeking and receiving mental health services, which will be discussed later in this chapter. One participant discussed the pressures of time constraints:

> I always feel so overwhelmed. I often feel I am on the edge of tears. There just isn't enough time in the day and I stress myself out trying to make sure everything gets done.

Another participant identified how the struggle of time for treatments was also being compounded by her child reaching an age where they want to be in control of their own care:

> Treatments are time consuming and my child is at an age where treatments are just a fight so it adds anger and frustration to an already inconvenient process. Now that she's starting school I am totally freaked out about how we will fit everything in the day and how she will adapt.

Limited Social and Familial Support

With time and money being the primary frustrations participants cited as influencing their anxiety, depression, and quality of life, lack of support was also discussed frequently. Struggles ranged from feeling as if participants needed to isolate (before the pandemic), to typical marital concerns, and lack of social friendships with those who understand life with CF. Marriage and parenting can be hard enough before taking into consideration raising a child with a chronic, life shortening illness, in the midst of a pandemic. Participants were open about their marital, familial, and social struggles. One mother discussed anxiety surrounding having differing parenting approaches or views on CF care:

> Differences in perception of risk between me and my husband and feeling he is risking our sons life exposing him to things I worry about due to cf or covid.

One participant, while not separating or divorcing, discussed the obstacles she was facing with her family:

> Kids fighting, not having any alone time, baby not sleeping, not leaving the house, not agreeing with husband about seriousness of Coronavirus and what to do about school.

One mother discussed her marriage ending and the difficulties associated:

> Fear of anyone in my family getting COVID, not being able to financially support my family, husband quitting his job and leaving us without health insurance, husband violating restraining order before divorce is finalized, husband dragging divorce out longer.

This mother reported the strain of raising a child with CF as becoming too much for her marriage to handle which ultimately led to its end:

> My son was born 11 weeks early. I had a great career that had just started and it abruptly stopped when he was born. I've never been able to get back to that career trajectory. My son was very sick early on and it caused a great deal of stress in the marriage. We tried getting pregnant using IVF and PGD and were unsuccessful so we tried the old fashioned way and had another child with CF. The stress of it all ended the marriage. I felt suffocated by medical issues and had no life outside of my kids. I love them like crazy but their dads life didn't change. Mine was. My life is dictated by CF. Consumed by it. every meal, contain carbs, dosing insulin, tracking blood sugars, counting out hundreds of pills. if someone suddenly popped into my life and had to do what I do on a daily basis, they'd probably go nutty. Luckily I've found a partner who is very supportive both financially and personally.

Another participant discussed similar struggles with a different approach:

> Covid has re-triggered a lot of my original fears and anxiety. We had our first in person clinic visit in 7 months last week and I was nervous leading up to it. We have also been displaced because of the virus. We left the city for the woods to isolate. My husband and I are separated and living apart but decide to leave together and live together temporarily to protect our daughter. He was unemployed and in the process of lining up a new job but then they announced they are on a hiring freeze. Although he made a good income, we now both have no money coming in except unemployment so this has caused more anxiety. We don't feel comfortable going back to the city. Finding a new home is hard without a job. We decided to rent something temporarily in a small town and homeschool the kids. Living together again is stressful and also re-triggering since he is one of my big triggers. he has not fully dealt with his ptsd/depression/anxiety and is prone to outbursts of anger.

While many openly discussed their marital struggles and parenting frustrations, some participants discussed feeling as if they had no support from friends or family and wishing for someone to simply understand their experiences as a caregiver raising a child who is chronically ill. Regarding changes in quality of life, one participant simply wrote:

> No help from family.

It is unclear if this was a change from prior to receiving a CF diagnosis, but I felt it important to include nonetheless. One mother replied to the question regarding depression contributions by heartbreakingly stating:

Mental health of my non CF child. Isolation and lack of friends who understand.

One mother reported:

> Working from [home] and not being able to go out and visit family and friends.

Another participant discussed lack of resources, support, and how CF care dominates her ability to receive support and how it has affected her marriage:

> We can't attend most family events. Going out is difficult due to enzymes. We can't see many friends. Few people can watch him. There is more fighting between me and my husband.

One father discussed how they changed their situation to be closer to receive familial support at the detriment to his educational and career plans:

> My wife and I moved from my dream-town, as I awaited my university to catch up so I could go to grad school. we moved back by her family to an area I only tolerate where I have to take whatever job I was offered, which at this point means a new car salesman, and settle for a house that is less than what I want; all to be by family support. I no longer look to the future more than a few days or weeks in advance between work and the CF appointments and treatments, and all it requires to keep my son "healthy". I feel overwhelmed enough as a new father, and then all of this happened.

Another mother shared similar feelings of daily overwhelm, lack of support, struggling to juggle other needs, and finding time for daily treatments as being her biggest anxiety contributions:

> Having zero help. Sometimes I just want someone to help with treatments that way I can do normal house duties and tend to my toddler. I constantly feel like I'm neglecting him and get easily frustrated because I just want to get treatment and meds done, but have to stop a million times to make sure my toddler is okay.

One mother discussed how CF has negatively impacted their ability to enjoy prior hobbies due to lack of support and concerns for her child's physical health:

> It has stopped all our family activities. We loved to ride dirt bikes and be out around the camp fire. We wanted our children to race motocross and clinic advised against those activities. I worked 8hrs a week to social and for extra income. I can no longer do that because no one is willing to watch my Cfer. We quit going to church for fear of our Cfer getting sick.

Overall Family Health and Safety

These parents also often battled with feelings of isolation due to fear of germ exposure and keeping their CF child as germ-free as possible in order to avoid potential devastating bacterial infections that could lead to them ending up requiring an unplanned hospitalization or worse. One father reported his struggles with anxiety around doing his part to keep his child physically (health) safe:

> I have severe anxiety about almost any kind of contact in the public world. This has sharply increased this year due to the Coronavirus pandemic. My anxiety is directly tied to exposure to cough, colds, and flu that I may pick up in the world and bring back home to my household. I take all the preventative care methods that I am able to; but I still have anxiety about every task that takes me out into

> the public. I worry about my son's health and what the future might look like for him; but have been able to mitigate that with meditation and other techniques which I have worked on with therapists.

One mother provided a short description of her current anxiety contributors:

> Concern over child's mental health and children's CF health. Concerned about Covid and finances.

It was apparent that not only were the physical and emotional concerns of participants focused on that of their child with CF, it appeared a majority realized the toll it could possibly take on other family members as well. Roughly 25% of participants directly mentioned either the physical or emotional health as a concern with an additional 40% discussing indirect health concerns such as their CF child's weight, PFT, or other health related issues of themselves or other family members. One mother stated her primary anxiety concerns:

> The mental health of my non CF child. Physical health of my husband. Physical health of my CF child. Fear of covid.

While another expressed similar concerns:

> I have anxiety about work, family relationships, financial stress from pandemic, empathy & stress from 3 extra foster kids in home, excessive worry about my cf daughters health and staying on top of preventative care, anxiety from my decision to homeschool even though my husband and I work full time, and feelings of inadequacy to protect my kids from the pandemic.

Other participants discussed health concerns without using phrases such as physical, emotional, or mental health:

> Child's condition deteriorating, cough [more] than usual and the fear of new bugs/ infections. Side effects of a lot of drugs - the team doesn't even care about informing us of potential side effects! Worry about living on single income and potential loss of job.

Pandemic Amplification of Distress

During the coding process, the coronavirus pandemic was mentioned by a majority of the participants and after careful consideration and an additional review of participant responses, by myself and second coders, it was determined the pandemic was highlighted in a way that required giving it a more thorough investigation. Therefore, "Pandemic Amplification of Distress" was the third theme to present from the findings, with two subthemes: (1) Physical Health Concerns and (2) Mental Health Concerns. The following section will highlight how the pandemic influenced this group's experiences with anxiety, depression, and quality of life, and how having a child with CF during a global pandemic further places the spotlight on their daily care and treatments to maintain their physical health needs.

Roughly 50% reported coronavirus as a contributing factor to their current anxiety, while 30% reported as influencing their depressive symptoms, and 20% as having an impact on their daily quality of life. The psychological strains of the coronavirus pandemic were plentiful ranging from feelings of overwhelm due to transitioning to working and learning from home, fear for the physical health of their CF

and non-CF children and themselves, employment (and lack of employment), isolation and being displaced, and general uncertainties regarding the pandemic.

One participant summed up many of the concerns of the participants that fit the main themes of the study:

> Covid, new baby, toddler with cf, not having family support available due to Covid, no regular outings I'm able to go to because of Covid. My angst currently is Covid related.

While another participant stated:

> Covid, back to school being immunocompromised, finances with hospital bills, and especially CF- not knowing what the next week will bring and what to do if it's bad all while arranging life around to accommodate.

The pandemic has led to an increase in anxiety and depression and a decrease in quality of life for many individuals worldwide. Individuals with CF are classified as being in a higher risk for COVID contraction, comorbidities, and complications, and it appears caregivers raising children with CF may be experiencing anxiety and depression related to the pandemic differently than others.

Physical Health Concerns

Many parents discussed their concerns of keeping their CF diagnosed child in a bubble versus letting them live a “normal” life. For some participants, the threat of exposure and contraction of the coronavirus increased those fears, leading to increased feelings of isolation and caregiver guilt. One father discussed his concerns for his son:

> The COVID pandemic and the concern about my son contracting the virus and not being able to successfully fight it off. This is a daily worry for me as we prepare for him to return to [retracted for privacy]. I worry about being the person who contracts COVID and give it to him. I worry that another member in the household contracts COVID and spreads it to him. I worry about him being in [retracted for privacy] and not getting the type of care needed if he becomes ill. I worry about losing my job and therefore my health insurance.

There were also concerns of their children contracting coronavirus and how that would impact their time in the hospital:

> Fearing sending the kids to school and having anxiety about letting them play with their neighborhood friends. Fear of my son being admitted & how it will be in the hospital during this pandemic.

Another mother expanded on the fear of her child ending up in the hospital and what would happen to him if something happened to her:

> Covid 19. I'm a single mom, if something happened to me, no one else could care for my CF child with autism and learning disabilities and my other autistic son who needs daily injection. I worry if my c.f. son got covid 19 how I would care for him at hospital and other son at home. I worry about disability insurance and medical coverage, I worry about restraining order for my ex, worry about finances.

Mental Health Concerns

Of the 81 participants who responded to the qualitative question relating to anxiety, 40 expressed the pandemic as a contributing factor. There was significant drop off in participant responses from question 2 to 3, and of the 57 responses to depression contributions, 17 discussed the pandemic directly. Interestingly, regarding quality of life, 58 participants responded, and 12 responded about the coronavirus influencing them currently. Much of the angst surrounding the coronavirus were related to feelings of uncertainty, the unknown, and feelings of loss of control. One participant expressed their fears:

> My children's health and future are the main cause of my anxiety. With COVID-19, the uncertainty of their well being makes me very uneasy.

Another discussed their concerns:

> Covid-19 and the whole uncertainty of the disease and the lack of solid treatment.

The uncertainty was problematic for many participants:

> Life has changed so much with COVID19. The concern and uncertainty cause me stress.

One mother provided insight into how the unknown was affecting her:

> It is slightly overwhelming having kids at home, navigating Covid, work and uncertainties.

Feelings of loss of control were found in participant responses such as:

> Little control over current conditions with COVID19 in our community.

The pandemic highlighted already present concerns:

> Covid, back to school being immunocompromised, finances with hospital Bill's, and especially CF- not knowing what the next week will bring and what to do if it's bad all while arranging life around to accommodate.

The coronavirus magnified the worries of many participants and the struggles they faced in their daily lives while also having to learn to pivot during the pandemic. A handful of individuals discussed how the isolation required was a struggle, not only for them, but their children as well. Accounts of COVID related frustration around isolation included:

> My anxiety is generalized and not tied to specific events, but Covid-19 and social isolation has definitely contributed to its increase. As a school teacher, the change has been particularly challenging.

One participant reported:

> State of the world, fear of my kiddo getting Covid-19, not being able to do anything.

Another discussed how the pandemic was bringing up past concerns:

> Mainly covid is the reason some feelings of depression are returning. Isolation from friends and family are hard combined with the worry of one of us getting it. Also I'm putting my career on hold for the next year to homeschool the kids and I'm worried about not getting the space I need for self-care.

One mother discussed how the pandemic isolation influenced her feelings of depression:

> Not severe depression but a feeling of hopelessness - that we have done everything we were told to do re: Covid but it is out if our hands. Other people

and the Gov now get to decide our fate and my son's health and safety. The feeling of being in control is gone.

Natural Skepticism and Cautious Optimism

While the daily care and treatment of the diagnosed child and CF as a whole were prevalent factors reported as contributing to caregiver anxiety, depression, and quality of life, many participants also identified the change in time from initial diagnosis to the present time as providing greater insight and awareness to their life with chronic illness needs and provided an opportunity to view their ability to transition and adapt as a form of resilience. On the other hand, many struggled with feelings of being continually overwhelmed and concerned for the future due to uncertainty. They questioned why this would happen to their child, how were they going to cope, and expressed beliefs that their situations would never improve. Thus, the fourth theme includes two related concepts: "Natural Skepticism" and "Cautious Optimism." These two concepts were present in the fifth experiential question regarding views on CFTR modulating medications, which will be highlighted within this section as a separate subtheme. A majority of responses highlighted participant insight, awareness, flexibility, adaptation, and resilience came through in the fourth qualitative question discussing how their child's CF diagnosis changed their quality of life past, present, and future.

Natural Skepticism

The skepticism many participants reported was generally related to feelings of disease care and treatment as well as concerns regarding the use of the CFTR modulating medications. Often times, feelings of helplessness due to lack of social, familial, and

financial support and hopelessness regarding CF physical health improvement were also present.

Additional narratives of natural skepticism came through more in the qualitative question regarding depression, rather than anxiety. One father reported his experiences with depression:

> I have struggled with depression since I was a teenager. My depression presents itself as a lack of energy, tiredness, numbness, and emotional emptiness. It can cause poor eating habits, sleeping too much, inactivity, and hopelessness. I notice that it comes in cycles, I sometimes experience depression for weeks or months at a time, and can also go days or weeks without it being an issue. Sometimes a challenge or difficulty in life can trigger a sudden depressive episode in me.

The feeling of skepticism, or even despair, was present throughout participant responses. It seemed as if they were unable to accept and believe in their current situations with joy or happiness due to a perceived cloud of cynicism based on their life circumstances leading to feelings of distrust toward self and others. One mother reported:

> I am not feeling depressed and have not been depressed for several years. I mostly feel on edge, like the other shoe is about to drop.

Another mother stated:

> Stuck feeling no way out ahead to move or progress while caring for cf child.

One mother reported her struggles:

> Being a good parent to my kids and partner to my husband seems impossible. It's almost like having to choose one over the other and it's crushing.

One participant discussed their worries:

> Feeling overwhelmed and hopeless. Feel like we are in a no win situation.

This mother expressed a continual feeling of self-doubt that had been plaguing her for years:

> I had a pretty difficult life before my little man was born, it' only gotten better, but more complicated. Things have improved financially, but the impending doom feeling has been here for the entire 8 years. I'm an artist (art teacher before I had [child]) and have felt inferior in my abilities lately. I took up sewing (to make masks) and it all feels pretty pointless. I have no suicidal ideations but do sometimes feel like running away. My therapist knows all this.

Finally, one mother provided a reply that touched on many of the narratives, themes, and categories presented here and in previous sections on how her child's CF diagnosis has changed her quality of life over time:

> This one was a little tricky. While I do feel overwhelmed or stressed about the daily life as a caregiver to a child with CF, I would never stop doing it. It's something I have to do, she's my daughter. It is frustrating that I feel the need that I've had to put my life (like working/having a career) on hold "in case something happens" Whenever I get to a place where I feel confident things will be okay, CF says "no." Which attributes to financial strain. We also have a pretty shitty support system. We have family and friends that say they "get it and want to help", but they never make the effort. Case in point, my fiancé and I have had a total of 3 nights without kids in 9 years. We haven't had a date in over a year. The

> fear of losing my daughter comes in waves. She relatively healthy, rather she is healthy. But I know one bad bacteria can quickly take that all away. It truly is soul crushing.

Another shared a similar experience:

> It's added a lot of stress and guilt to my life. When I feel like I can relax a little bit it seems like we get a new diagnosis or their health has changed. I go to bed feeling like a failure because I'm so overwhelmed I sometimes forget a medication or treatment. I don't have friends any more so other than my parents and a few coworkers, I don't socialize. I always feel like I'm a burden to others if I ask for help.

Cautious Optimism

The first half of this section may be discouraging and disheartening to read for some. However, it seemed that for every skeptical response there was an opposite optimistic response as they recognized their strengths as a resilience resource to adapt to the obstacles they had been presented. One father highlighted how his family learned to pivot along with the challenges they face daily:

> In the positive realm his diagnosis has provided a clarity of being present and enjoying every moment. We value every day we have together. It also comes with a lot of responsibility of doing treatments and taking medications multiple times per day. That is a big commitment for a child, and equally so for us as parents. We do limit what we are comfortable doing social; we avoid crowded stores, movie theaters, restaurants, large auditoriums or stadiums. We homeschool to

> avoid the exposures and risks in classrooms. Despite those issues, we still find ways to have an active life, with outdoor activities, socializing, and have fun every day.

Another mother discussed how her recognition of needing a shift from negative to positive has enhanced their lives and adjustments they make to prioritize health for all:

> After years of wallowing and depression, we learned to be positive and embrace the life and time we have. Being stressed and negative everyday was no life to live. Health is the biggest factor. Extreme CF fatigue means no walking long distance. Too much heat sets off fatigue. Environment contributes. We stay out of smoky and Dusty places or if feeling well, will use a dust mask out and about.

The idea of slowing down, being in the moment was discussed by various participants in responses such as:

> Cf dx [diagnosis] has added more emotional and financial stress to our family. It has also caused us to slow down and enjoy life in the moment.

For some, their journey into the CF community led to inspired purpose in learning advocacy, awareness, and fundraising for the CFF in the hopes of finding a cure. One mother discusses her journey from early diagnosis 12 years ago to her relationship with CF today:

> My child having CF has changed my life. When she was first diagnosed I went through all kinds of emotions, especially extreme anxiety and depression. I was terrified for her life and well being and with administering all her treatments. It eventually got easier, but my focus was either on keeping her healthy (daily

> treatments) trying to get her over any sort of cold, tune-ups and surgery would cause everything in my life to stop while these things were going on. I became obsessed with her lung function, breathing sounds, and stamina levels, right along with the color of her mucous. And when I wasn't obsessing over these things I was trying to find ways to raise money for CF research so she had a future. Now that she is 12 and on Trikafta I feel she has more of a future, but there are so many unknowns with Covid-19 and the effects on her body from being on a drug like this long term. Trikafta has removed a great deal of my worry allowing her and I a better quality of life.

Another mother discussed a similar trajectory while also acknowledging the emotional toll it has taken on her:

> I did not envision that my life would require such limitations having a child with a chronic disease. Before my child was born and diagnosed, I was much more hopeful and peaceful. I am now more anxious. However, my child's diagnosis changed my focus completely and how I work in advocacy and healthcare improvement. I am hopeful my child will eventually take over control of their disease. When this happens, I will be able to rest some. Although a parent of a child with CF is always a parent of a child with CF. The daily workload may improve but the anxiety likely to continue throughout life on some level.

One mother expressed how her life had changed and the struggles she faced while ultimately recognizing her time with her child is the most valuable to her:

> I can't work since he requires so much. My work insurance wouldn't cover the expenses, which leaves us in the poverty/disability cycle. I miss working and feeling independent. I don't love the stigma that comes from being on disability. I do appreciate how much time I have with my cfer. I know I won't ever regret our time.

One father participant discussed his struggles and how the pandemic has influenced their home life while also expressing hope for his son's future:

> My son's CF diagnosis was like a kick to the stomach. It changed everything and nothing. The daily protocol was extensive and required major routine changes at all levels of my life. Currently we are on a great routine, but COVID has put a wrench in our routine. I am hopeful for the future. With a vaccine, I can see my son returning back to his normal routine and live a great life. My greatest fear continues to be that he becomes ill from this pandemic.

Finally, one mother reported in an easy-going manner how they have adapted to their circumstances and what they do to maintain their status quo:

> Covid-19 changed our lives. We carry on. She is healthy. We bike, I scoot in a chair. We built a treehouse. We have finally returned to church. Our daughter and family live next door. My mom in law on other side. My sister best to be. We see my dad every Sunday. We work on cars. We stay busy.

Skepticism and Optimism Toward CFTR Gene Modulating Medications

A total of 56 participants made it to the end of the 14-page survey and answered the fifth qualitative question regarding their perspective on the CFTR gene modulation

medications. Participants were prompted by the statement “Many individuals within the CF community express their hope and excitement with the introductions of the CFTR modulator medications including: Kalydeco, Orkambi, Symdeko, or Trikafta.” They were then asked if the introductions of these medications had changed their viewpoint, perspective, or relationship with CF, and to describe how their thoughts and feelings had changed or evolved regarding CF in their current day to day life and going into the future. Only one participant reported they had not heard of any of the medications or their availability. A majority 75% of participants expressed their excitement, hope, and encouragement at the availability of these medications, especially Trikafta. Forward focused positive emotions were expressed as caregivers discussed they felt less worry for their child’s future through physical health and normalcy and also looking at longer, brighter, and healthier lives for their children. One participant went as far as saying:

> There's no better time to be born with CF!

One mother believed the introduction of these medications to be so powerful, she has changed her outlook on the disease as being life-shortening:

> I feel like their future is much brighter. They have more energy on the modulators. They also have more hope. The CF community is so much more optimistic than 20 years ago. We see CF as a chronic disease and I don't even like to say it is fatal.

Less than 15% admitted they had concerns surrounding the lack of data and long-term side effects. One mother expressed defeat:

> I was excited until I started hearing about liver failure because of it. You can't win.

Another mother remained hesitant but was staying focused on the positive:

> More hopeful. Still leery of long term effects of modulators, but excited nonetheless.

One mother who piloted the survey expressed hope, decreased worry for her daughter in general, and also identified concerns of long term side effects:

> Since she has been on Trikafta and showed tremendous improvement in her lung function, I have worried less about her future. I worry less that she will need a tune up and miss out on school and life, I have worried less that she will be less likely to participate fully in all activities, especially cheer. I feel that she has a bright future and I feel positive that pretty soon she will need less and less of all the other medication she is on. Although I still worry about the long term effects the new medication will have on her, I worry about her liver function. But the worry is very different from the worry I used to carry around. I don't worry as much if she misses a treatment here or there anymore. And it is great to see her feeling better and healthier.

Two participants reported their children were taking one of the medications previously but had to stop due to complications. One parent expressed her sadness:

> Had such high hopes. Yet, they keep making his liver counts skyrocket & we have to discontinue. Disappointing.

One mother discussed her daughter's experience:

> Kalydeco was a horrible experience for my daughter. She was far worse on it. She not only had physical side effects, but it caused severe sadness and anxiety. The only good that came from what was she was less salty so she was taken off after three months. Not worth it in her case. The terrible side effects is something that needs to be discussed more.

Approximately 30% of participants reported their child did not have access to these medications due to age restrictions, CFTR gene mutation, disease severity, liver functioning, or the cost of the prescription drugs. Approximately 10% of the CF population is not currently eligible for modulators due to gene mutation restrictions, but this participant remains hopeful that a modulator will become available in the future for their child:

> Still holding hope for the 10% as my child is part of them.

This mother also expressed hope that eventually her child would also be included:

> I feel hopeful that eventually a CFTR modulator will be developed that is targeted for his mutations.

The inability to access the medications due to her daughter's current liver functioning is devastating to this mother, but she remained hopeful:

> I have been hopeful for better, longer lives of those with CF. My daughter has been told she can't use any of the current modulators due to the severity of her liver disease. That has left me scared that there may not be a solution to help her CF.

Another mother discussed lack of clarity on availability due to her son's liver function:

> It's still not clear if my son can take Trikafta as he has severe liver disease.

Some of the participant's children were currently on one of the medications and almost 20% stated their child was showing significant health improvements since beginning their medication. One account of how Trikafta was benefiting their child:

> Trikafta is life changing for [my son]. He has been in and out of the hospital a lot the last two years. Trikafta has improved his lung function and cleared bacteria from his lungs.

One participant's child was seeing great results with Orkambi:

> I have noticed a significant improvement in my child's health since she has been on the Orkambi. I am super excited to see what new medications come out in the future. I see a long life in my child's future.

Although this mother is seeing results in her son while on Kalydeco, she was still hesitant to trust the process fully:

> I feel the Kalydeco has helps my son a lot. Concerned about long term side effects. Everyone thinks they are so great and it will be a cure all but I am still very leary.

Only one participant had a singular experience with Symdeko and reported:

> Since we saw no change when starting Symdeko, I don't think any differently about cf.

However, one participant's child had been on multiple modulating medications and expressed her excitement and satisfaction at the experiences of each new drug:

> My daughter started Kalydeco in 2013 and it was an absolute miracle drug for her. She had gotten so sick and her health was declining fast until she started Kalydeco. In 2019 she started on Symdeko to see if it would work any better for her and it did. She gained more weight than she ever had (one of her biggest struggles before was being very underweight). Just within a few months, Trikafta was approved so she switched to that and even more progress was made with her health since then. She's doing better than she ever has. Each drug just keeps getting better and better.

One mother summed up her feelings of both optimism and skepticism:

> I'm hopeful that these drugs will greatly increase the quality of life. I'm hesitant to jump on board right away because she is currently doing so well And I'd like to see how patients do in the long run. I tend to distrust pharmaceuticals and the western medical model so I come from a different mindset. I focus on making sure she has a well rounded healthy diet, exercise, massage, and other complimentary healing modalities which seems to be working for her. However, I know that these meds can be life changing and I'm thankful they are there if and when she needs them. Thankfully she is a very positive and joyful child and easily does whatever she has to do to stay healthy. This gives me a lot of reassurance that she will be ok in the future with or without the advances in science.

Two fathers made it to the end of the survey, and they both touched on the topic of hopefulness for their children and this population. One father reported:

> Ever since we started taking the CFTR medications (started in Jan 2018) he has had little to no issues will illness, cough, or colds. It has really given us hope that he will an easier time with the treatments and medications in the future, than what was available to previous generations of the CF community.

Another father stated simply:

> We have always been hopeful. But since the medication we're extremely hopeful she will continue to get better.

Barriers to Seeking and Receiving Mental Health Services

This section includes barriers identified by the participants in their responses to the first experiential question: "If you have considered seeking mental health services, please identify specific barriers to seeking and receiving mental health treatment for yourself" and is the fifth and final theme. An overarching goal of this study was to help bridge the gap in literature to help continue the dialogue of best practices to serve this population; solutions to problems cannot be found without also acknowledging how individuals come to find themselves with the problem in the first place. Four subthemes were identified as barriers affecting a caregiver's ability to seek services including: (1) No time or money, insurance or support, (2) Mental health stigma, (3) Access to mental health care professionals, and (4) Natural skepticism toward mental health care. Mental health stigma and access to mental health professionals were two prominent subthemes that emerged from the data, with over 25% of participants identifying stigma as a contributing factor and approximately 20% discussing the lack of access to therapists as a barrier.

No Time or Money, Insurance or Support

A majority of the 82 participants who responded to this question reported that a primary barrier to seeking and receiving mental health services was lack of resources (time, money, insurance) and lack of support (childcare and familial support, inability to take time off of work). When responding to barriers to seeking mental health care, one mother reported frankly:

> My only goal right now is keeping her alive.

Another mother stated her biggest cause of anxiety:

> My CF son's life. How to keep him alive as long as possible.

One mother identified also struggling with panic stating:

> This disease is terrifying.

Another mother identified her struggles as follows:

> Going to a lot more doctor appointments then I ever have, more than the other family members combined, having a constant worry of my child getting sick, child missing school, hospital stays and missing out on activities. Currently with pandemic, more worrying, more missing out on things, especially friends and school. My son has a gtube and having to do feeds throughout the nights, waking to alarms, having to ensure he is fed correctly and enough to take medications (Kalydeco), wishing he would learn to swallow pills, worrying if he will be able to have children naturally, worrying he will get to do the things he wants to do in life. My child has recurrent pancreatitis and whether or not he has pain during the day or night contributes to our quality of life.

Almost half of the 82 participants who answered this question identified time as being a contributing barrier to receiving mental health services. Finances and lack of childcare were also discussed by many participants. One participant reported:

> Lack of time. Already spend too much time in hospitals and clinics. Too many appointments.

Another stated:

> I cannot fit an appointment into my work day. Plus any extra leave/vacation time at work is used for doctors appointments and hospital trips. I'd like to focus on my mental health to better care for my CFer but I had a lot of childhood trauma and PTSD so I feel like it opens a huge can of worms and emotions that I don't have time to process.

Many participants kept their responses short, sweet, and to the point, citing "lack of funds," "lack of time, finances, and childcare," "mental health, insurance, coverage for meds," and "lack of access" as contributing barriers. Others elaborated a bit more providing some insight into their specific barriers:

> CF care itself takes up a lot of time, so haven't been able to prioritize own mental health. There is a psychologist at the clinic but going to clinic with an anxious child who is complaining and crying itself is very stressful and the clinic is not very understanding either, so haven't asked for their service so far as the idea is to get out of the clinic asap.

One mother reported:

> With the kids home I cannot go to my doctor appointments. My husband works &

> I have no family close. I have anxiety and the pandemic is definitely making it worse along with depression but the appts are all in person and I can’t take my children.

One of the study’s fathers shared his emotionally devastating experience for himself and his daughter when discussing barriers to seek and receive mental health services:

> I've considered it but being a single parent 24 hour 365 days a year caregiver to the most awesome daughter a father could ask for leaves very little time for me to be concerned about myself. My daughter is in the late stages of CF and her Dr has told us there wasn't anything else he could do for her. This has psychologically been a disaster for her. She started having severe panic attacks, depression, and in February of this year she almost gave up as she told me. Being the warrior she is survived. One week ago I had to call 911 as her 02 canula fell off and before I could bend over, pick it up and hand it back to her she had become unresponsive it took a lot of 02 to bring her back around. I'm a nervous wreck , sleep deprived, and worried to death. I know my [daughter] cannot survive without supplemental O2. I have trouble sleeping due to worrying about a power outage. This is the hardest thing I’ve dealt with in my life. I truly don't know how I do it, love for her is a powerful motivator.

For a population already bogged down with two to four hours of daily CF care and treatments, quarterly clinic visits, and more frequent tendency for hospitalization, it is not surprising to read that the resources of time, money, insurance and lack of support, either

from family, friends, or work would be the top contributing barriers to seeking care for themselves.

Mental Health Stigma

In reading participant responses, mental health stigma as a whole was present, including both internalized and externalized beliefs. Twenty-five percent of participants identified mental health stigma as a barrier to seeing a therapist or counselor. Corrigan and Bink (2016) identified four types of stigma: public, self, structural, and label avoidance. It was apparent that participants were experiencing public, self, and label avoidance stigmas within their responses. The implications of mental health stigmas will be addressed in greater detail in Chapter 5. An example of a participant's self-stigma and label avoidance:

> In the last 3 years I've considered seeking mental health lots of times, but I'm worried my doctor would think I'm unable to cope. I have good days & bad days, but I class myself as unimportant as my CF child is my main priority.

An example of public, self, and label avoidance stigma can be seen in one participant's response and fear of potential court proceedings:

> [I] don't want to appear unstable in court if there were ever a custody battle.

Many participants had responses that fell into the self-stigma and label avoidance category. They identified themselves as feeling selfish, guilty, and embarrassed as barriers while others reported:

> I am not a priority.

Some were too exhausted:

> I have no energy left to focus on self.

Others felt as if they didn't take center stage:

> I put myself on the back burner.

Internalized fears held others back:

> I have fears of seeming inadequate.

Worry of what others would think:

> I have fears of being judged.

One acknowledged not putting themselves first:

> I just neglect myself.

Many cited a general feeling of fear of the therapeutic process and being emotionally vulnerable and also being fearful they would be seen as being unable to take care of themselves and their children. One participant reported:

> Time, childcare, always making sure everything is done for everyone else, and fears of seeming like I can't handle taking care of my kids if diagnosed with any mental illness issues.

Another mother reported:

> I would never say in front of my child with CF, that would be selfish, he has to deal with it so me feeling bad is selfish and I wouldn't want him to find out.

Access to Mental Health Care Professionals

This subtheme includes many facets of "access" to therapists and counselors including therapist fit, their knowledge of CF, geographic location, scheduling, being

overwhelmed by the process of finding a therapist, appointment wait times, and uncertainty of the therapeutic process as a whole. Fifteen percent of participants discussed struggles with access to a therapist. One participant reported a physical access barrier stating the location being 6 hours away as being problematic. Unfortunately, it is unknown if this individual lives in an incredibly rural area or if they meant specifically their CF clinic was 6 hours away. Nevertheless, it is important to address this finding as access to therapists in rural communities can be a difficult process for various reasons (Rural Health Information Hub, 2021).

Additional barriers within this subtheme included concerns of not knowing where to start, lack of clarity with insurance, long wait times, being uncertain it would be helpful or effective, and feeling as if medication would be the only answer or option. The most common response was regarding lack of therapist knowledge of CF. One participant reported their struggles:

> Lack of time, unable to leave kids alone to drive to and from and spend an hour at a clinic, I feel I have to explain CF over and over again every time I meet a new therapist, finding a therapist nearby, and cost.

This response highlights all of the main themes except the pandemic. One father replied:

> The largest problem has been availability of therapist[s] who are familiar with CF and the CF complications. In Austin, Texas we found a great therapist who worked to learn and educate himself on CF and its unique problems. In Lawrence Kansas we have yet to find a therapist willing to learn the unique issues facing a CF patient and the associated issues while dealing with transitioning to self-care.

Regarding insurance coverage/confusion:

> Kaiser does not have a mental health department. This department is vague and limited. I have asked several times to speak to someone and always get the 'okay we will have a social worker call you.' It's been 9 months and I still have yet to speak to anyone.

Another participant discussed numerous barriers:

> Even with great insurance counselors are hard to arrange, appointments are often two to three weeks out, not ideal for an anxious person.

It is important to note that not all individual's experiences barriers to seeking to services. A few participants were actively receiving services, reported having great insurance coverage, and acknowledged there were no prominent barriers to receiving care. A handful of participants stated they had not considered mental health services, were not currently in need of a counselor or therapist or preferred to manage their mental health on their own. Surprisingly, only three participants had mentioned the Coronavirus pandemic as a barrier to receiving treatment.

Natural Skepticism Toward Mental Health Care

While the majority of this section was focused on the biopsychosocial factors surrounding the caregiver's experiences in raising a child with CF and its challenges, it is important to also quickly highlight the natural skepticism regarding the mental health process. A few participants reported their experiences with past attempts at therapeutic treatment and the responses provided fell into the natural skepticism subtheme. One participant reported their barriers:

> Money guilt exhaustion. I don't know who to talk to. I tried one counselor and I think he never understood me. It seems pointless to try to explain to a counselor.

Another stated:

> Distance of providers, lack, tried several times and have not found right fit, overwhelmed with how much work it is.

One mother's response sounded as if she was interested in seeking services, but identified her barriers, and included her perspective on her husband's lack of seeking treatment as well:

> Cost is a barrier for me. For my husband, he won't access mental health treatment and I think this may be due to inhibitions.

Finally, one participant was brutally honest with their experience leading to disclosing blatant feelings of skepticism toward therapy:

> I have tried to speak to a therapist over Telehealth. It was ineffective and a waste of time and resources.

Summary

This chapter discussed the methodological approach for the study including sampling and recruitment, procedures, quantitative instrument information, qualitative survey questions, research questions and hypotheses, statistical analysis selection of MANOVA, child and caregiver variable tables of measurement, and the reason for utilizing a phenomenological genre background. Unfortunately, none of the quantitative research hypotheses were able to be accepted and retained due to the overall lack of significant findings, although a few univariate results were found to be significant.

Five overall qualitative themes and subsequent subthemes were identified. The first theme "The CF Priority" consists of Daily CF Concerns, Prevention of Spontaneity, Balancing CF Care Needs, and Grief and Ambiguous Loss subthemes. The second theme, "Resources and Support," had subthemes of Time and Money, Limited Social and Familial Support, and Overall Family Health and Safety. The third theme, "Pandemic Amplification of Distress" had subthemes of Physical Health Concerns and Mental Health Concerns. The fourth theme comprised two similar concepts, "Natural Skepticism" and "Cautious Optimism" with a subtheme of Skepticism and Optimism Toward CFTR Gene Modulating Medications. The fifth theme, "Barriers to Seeking and Receiving Mental Health Services" had four subthemes including No Time or Money, Insurance or Support, Mental Health Stigma, Access to Mental Health Care Professionals, and Natural Skepticism Toward Mental Health Care.

CHAPTER V

IMPLICATIONS, RECOMMENDATIONS, AND CONCLUSIONS

Discussion

The purpose of this study was to understand the lived experiences of caregivers raising a child with cystic fibrosis in relation to rates of anxiety, depression, and quality of life, and to answer the question, "How do biopsychological factors contribute to elevated rates of anxiety and depression, and lower rates of quality of life for caregivers who are raising a child with a cystic fibrosis diagnosis?" A mixed methods study was conducted and worked to uncover the experiences of 155 parents raising children with CF. To expand on the work of the TIDES study (Quittner et al., 2014), quantitative components to measure participant reports of anxiety, depression, and quality of life with the online assessments GAD-7 (Spitzer et al., 2006), PHQ-9 (Kroenke et al., 2001), and CQOLCF (Boling et al., 2003), respectively, were included. This study sought to expand upon the TIDES data, on a smaller scale, and to also incorporate the contextual element of experiential questioning. The parents in this current study also completed the CFQ-R (Quittner et al., 2005), a measure of the quality of life of their child diagnosed with CF as perceived and reported by the caregiver. The FSGI Model (Rolland & Williams, 2005) provided the theoretical foundation for this research.

There are about 30,000 individuals currently diagnosed with CF in the US, with roughly 1,000 new cases diagnosed annually, and around 70–100,000 cases worldwide, making CF the most common genetic, chronic, and life-shortening disease of Caucasians

(CFF, 2014b; CFWW, 2019). This is a disease that has over 1,700 variations making it difficult to pinpoint a generalizable treatment plan for all affected (CFF, 2014b), with a high treatment burden consisting of, on average, 4 hours of daily care required (Sawicki et al., 2009), and a predicted survival rate of 46 years or older for approximately half for those born between 2015 and 2019 (CFF, 2021a).

Quantitative Review

How Age and Sex of the Child Influence Caregiver's Emotional Health

Although the research question and alternate hypothesis for this section was rejected, the overall MANOVA model for child age group was significant, with participants experiencing higher levels of anxiety and depression when raising children in the 0–6 years age group. Caregivers of boys and girls ages 0–6 years old reported significantly higher, similar levels of anxiety when compared to other age groups of both boys and girls. Caregivers of male children reported significantly higher levels of anxiety when compared to caregivers of female children. However, the most important finding was the between group differences for caregivers of female adolescents ages 14 and older who reported the lowest rates of anxiety and depression as well as the highest levels of quality of life when compared to parents raising males and all other age groups. The TIDES study also found that raising a younger child was a contributing factor to mother's rates of anxiety, however the age and sex of the child did not affect father's rates of anxiety, or rates of depression for caregivers (Quittner et al., 2014).

This data tells us that caregivers may need additional increased support in the early years as they receive their child's diagnosis then need to learn how to care for the

child specific to their CF needs, while also trying to adapt, and cope with their new circumstances getting familiar with the CF community and all it encompasses. It is unknown why caregivers of children ages 0–6 experienced higher rates of anxiety and depression than any other age group. None of the participant responses indicated any noticeable differences in their qualitative responses to help better understand the correlation. The 0–6 age group had the lowest representation with the highest impact and that may also be due to the first years of parenthood being filled with the most change.

The data also brings into question how raising an adolescent daughter differs so drastically and appears to have minimal impact on the emotional health of the caregiver when compared to raising adolescent sons or children in the other age groups. Within this current study, male children outnumbered female children almost 2:1. It may also be important to note that children in the 7–13 group had the greatest representation while the 0–6 and 14 and older categories were almost evenly split, minimizing the potential for a skew in data distribution. An assumption could be made that adolescent females have less of an emotional impact on their parents due to the social construct of girls being perceived as being more mature, and potentially taking responsibility for their treatments compared to their male counterparts.

It is important to point out that the alternate hypothesis was primarily rejected based on its wording: “caregivers with infants and female adolescent children will have higher scores of anxiety and depression and lower scores of quality of life,” and grouping each of the emotional health indicators into one category. Despite the lack of overall statistical significance for the model, the significant univariate findings in this current

study suggest agreement that parents raising children from 0-6 experience higher rates of anxiety and depression and lower quality of life. Additionally, parents raising adolescent females experienced higher levels of quality of life and the lowest reported rates of anxiety and depression. Interestingly, a recent study by Cronly et al. (2019) found no association between the age or gender of the child and parental rates of anxiety and depression.

The Female and Male Caregiving Emotional Health Difference

According to the TIDES data, rates of anxiety and depression within the CF population are two to three times higher than that of the general population (Quittner et al., 2014). The TIDES study reported elevated rates of depression in 37% of mothers and 31% of fathers, and elevated rates of anxiety in 48% of mothers and 36% of fathers within their sample population (Quittner et al., 2014). The rates of depression reported in the TIDES study are three times that of a community sample, and anxiety rates reported were two to three times higher than those reported in community samples (Quittner et al., 2014). In this current study, elevated rates of anxiety and depression were measured using the cutoff point of moderate indication (greater than or equal to 10 and above) to severe. Fifty percent of mothers and 34% of fathers indicated experiencing symptoms of depression and 59% of mothers and 44% of father reported symptoms of anxiety. A side-by-side comparison of the TIDES data and this current study may be found in Table 23.

Table 23

TIDES Study versus Current Study Caregiver Reports of Anxiety and Depression

Emotional health indicator	TIDES (%)	Current study (%)
Depression Rate		
Mother	37	50
Father	31	34
Anxiety Rate		
Mother	48	59
Father	36	44

Overall, while only 28% of participants identified as being currently diagnosed with depression, their scores on the PHQ-9 identified 48% as experiencing symptoms indicative of moderate to severe levels of depression. Similarly, participant GAD-7 scores indicate 57% were experiencing moderate to severe levels of anxiety at the time of the study with only 45% reporting a current anxiety diagnosis. These numbers indicate approximately 20% of the participants had undiagnosed depression and 12% had undiagnosed anxiety.

This current study found similar results to Driscoll et al. (2009), with fathers reporting higher anxious symptoms compared to depressive symptoms as well as higher scores of quality of life than mothers. What we also observe, is that as a whole, a majority of this group is struggling with mild to severe anxious and depressive symptomology and could benefit from further evaluation and clinical therapeutic support or psychotropic medication. This current study also shows caregivers are experiencing higher levels of anxiety and depression than previously reported in the TIDES data, which may be related to the impact of the coronavirus pandemic, which is discussed in greater detail later in this chapter.

The Impact of CF Gene Mutation and CF Severity on Caregiver Emotional Health

The hypothesis that the child's CF severity and CFTR genetic mutation would negatively impact caregiver rates of anxiety, depression, and quality of life was rejected. It may appear that CF severity and genetic mutation do not play a role in the emotional health of caregivers. However, the parents in this study were primarily raising children with mild to moderate CF severity and a F508del mutation. Due to the lack of variety in types of CFTR genetic mutations in this study, it is likely that the sample sizes for the less common mutations were too small to reach significance. As highlighted in the results section of chapter four, there were very minimal differences in parental rates of anxiety, depression, and quality of life with reported rates showing as very similar regardless of CF severity or genetic mutation. The data suggests these caregivers were experiencing a moderate level of anxiety and depression despite their child's mutation or disease severity. Quality of life, reported overall was average, ranging from 70 to 75 on a scale that gauges a high quality of life target number of 140. This could potentially be explained by the increased levels of anxiety and depression in general, the amplification of pandemic distress playing a role, or perhaps raising a child with a chronic illness is equally as stressful regardless of the perceived or actual diagnosed disease severity. It is interesting and should be noted that although rates were similar with little differences, the caregivers raising children with severe CF reported the lowest rates of anxiety and depression, and highest quality of life when compared to those raising children with mild or moderate CF. This may be a reflection of the distribution of participants as a majority

reported they were raising children with mild to moderate CF, and only 18 of the 117 children were reported to have severe CF.

There is conflicting data regarding the relationship between CF severity and parental emotional health. One study found parental quality of life was negatively impacted as their child's CF severity increased, measured by their lung function; (Driscoll et al., 2009). Alternately, Besier and Goldebeck (2011) found no association between the physical health of a child diagnosed with CF and the mental health of their parents, when looking at adolescent-parent dyad relationships. More research is warranted looking at CF severity as the data is conflicting. Additionally, looking at the relationship between child CFTR genetic mutation and the mental health of the diagnosed child's parent caregivers was unable to be located. What the data of this current study does support is that while 18% of participants had a child with severe CF, 37% had moderate CF, and almost 48% had mild CF – this is a resilient group who adapt to their circumstances and needs of their child regardless of their mutations or disease severity without influencing their overall quality of life, anxiety, or depression.

Does CF Care Clinic Distance Matter?

It was hypothesized that a caregiver's distance from their child's CF clinic and availability of in-person support and community would negatively impact rates of anxiety, depression, and quality of life. Research looking at the relationship between parental mental health and CF clinic distance was unable to be located. In this study, the distance alone to CF clinics was not a factor in the participant's emotional health. This may be due in part because they have accepted that they need to carve out time to go to

clinic a minimum of four times per year and have made that commitment. It could be that those who are farther away from clinics utilize virtual communication; many participants reported they had the opportunity to text/call/email their providers before the world moved to the virtual format, and quite a few identified this option as being newly introduced in light of the pandemic.

The lack of significant findings may also be limited due to creating three categories of distance for running the MANOVA rather than testing based on a continuous scale. In future studies, this aspect could be analyzed in a different manner potentially resulting in different results. The time and distance commitments appear to be a moot point for many parents because they recognize that without these clinic visits their child's physical health could deteriorate, their life could be shortened, and the quality of their lives could also diminish. This is a group of parents that are dedicated and take very seriously their role of extending their children's lives in any manner possible.

Qualitative Review

The emotional toll was enormous on the parents providing daily care for their children with cystic fibrosis along with the potential for increasing disease severity and negative trajectory of the disease. The five themes yielded from the qualitative coding: The CF Priority, Resources and Support, Pandemic Amplification of Distress, Natural Skepticism and Cautious Optimism, and Barriers to Seeking and Receiving Mental Health Services, helped to provide insight into the struggles of this population. From parents and families being unable to be spontaneous and just pick up and go, to the challenges of finding time for daily CF care and treatments, as well as routine hospital

visits, doctor's appointments, and clinic visits – the physical, mental, and emotional burden this disease requires is worrisome.

Both themes, The CF Priority and Resources and Support, reinforced the sacrifices that the families made to ensure the best care for their child. The cost of medications and care alone were extraordinary. Due to the overwhelming cost of treatments, therapies, and medications, it is common for almost half of individuals with cystic fibrosis to receive some form of Medicaid and be eligible to receive Social Security benefits (CFF, 2020b). According to the CFF (2020b), diagnosed individuals must seek medical care at specialized medical facilities (the CF Centers), and it is common for a household with a CF diagnosed individual to pay over $10,000 annually for premium costs, and that doesn't take into consideration insurance copays, coinsurance or noncovered expenses. Individuals with CF (and their families) may utilize the CFF Compass program to help them better understand insurance options and navigate legal and financial resources related to disease management (CFF, 2021b). The CFTR gene modulating medications alone have exorbitant annual costs ranging from $268,963 to $311,501 (House, 2019; Martins, 2019; Starner, 2019). A breakdown of these drugs, all manufactured by Vertex Pharmaceuticals, can be found in Table 24.

Table 24

Cystic Fibrosis Gene Modulating Medication Annual Cost

Drug	Annual Cost ($)
Orkambi	268,963
Symdeco	288,000
Symdeko	307,238
Trikafta	311,501

The Pandemic's Impact on Emotional Health

While the pandemic presents challenges of its own for everyone around the world, it was not surprising this issue was highlighted throughout the survey. The coronavirus is an airborne respiratory illness and these caregivers are raising children who have a disease primarily affecting the respiratory systems, including the lungs and mucus production. It appears the coronavirus and its accompanying pandemic had a prominent influence on caregiver's anxiety, depression, and quality of life. It incited even more fear, worry, and concern than may be observed in the general well population to the combination of the COVID disease symptoms as well as the underlying conditions of living with CF. As mentioned above, this current study's participants had even higher elevated rates of anxiety and depression than reported in the TIDES study, which were already two to three times higher than the well community sample (Quittner et al., 2014). The increase in this current study's reported levels of anxiety and depression could be explained in part by the amplifications of distress from the coronavirus as parents attempted to navigate their lives around the uncertainty of the pandemic, while also raising a child who falls into the higher risk category.

Ambiguous Loss

When discussing participant's natural skepticism and cautious optimism, more was learned about their mental health struggles, especially grief and ambiguous loss. While many participants discussed grief directly, many of their responses fell into the category of ambiguous loss. This type of loss is an atypical family theory categorized as a relational disorder through psychological distress where loss is unclear, uncertain, and

based on ambiguity and lack of information/clarity within relationships (Boss, 2007). A tangible example would be caring for an individual who has progressive Alzheimer's disease: the individual you love is physically present, but mentally and emotionally absent. Therefore, there is no closure available, it is impossible, and as such one is unable to appropriately grieve their circumstances; thus, they are stuck in an emotionally frozen state and may view their situation as traumatizing (Boss, 2007).

A core assumption of ambiguous loss is the resilience of the family and how they cope through trauma when they are frozen in grief. Those experiencing this type of loss work to move forward with their lives while carrying their unanswered questions and trying to create meaning from their situation. For caregivers within the CF community, there were many unanswered questions, some stated and others not. The lack of control, uncertainty, and unknowns were all contributing factors to participants anxiety, depression, and perceived quality of life. However, what participants expressed within their responses were those on a resilience spectrum, if you will. There were participants who had not yet created meaning from their child's CF diagnosis and were stuck in a cycle of feeling as if they were in a space of "impending doom" or "waiting for the other shoe to drop," and even questioning why God would do this to a child (less coping and resilience). Other participants were in the middle, toggling somewhere between creating meaning and learning to make space for themselves by finding balance between the daily routines of CF and living a "normal life," while also still expressing natural skepticism of their child's ability to live a long, healthy life and worry for long term physical health concerns. Then there were participants who were on the opposite end of the spectrum;

those who had embraced their lives with CF with cautious optimism, who were attempting to make the best of their situations and were learning to be more present and appreciative of their circumstances, recognizing they would not change their parenting option if given the chance.

The Stigma of Mental Health

Participants shared their experiences of barriers to seeking and receiving mental health care. A discussion of stigma is important to acknowledge, as there are oftentimes misconceptions of how stigma may be perceived or applied. Participants in this study discussed concerns that they would be judged by others, viewed as being unable to care for their children, and not wanting to appear unstable if ever needing to go to court. They voiced hesitation towards receiving a mental health diagnosis and/or being prescribed medication. They also reported feeling selfish, guilty, and not important enough to put their emotional health needs as a priority.

A brief overview of the types of stigma include social (public) stigma, whereby individuals are negatively viewed, prejudiced, or discriminated against based on socially constructed ideals by the larger population (Corrigan et al., 2014). Self-stigma is when the overall negative beliefs and attitudes of the general population (social stigma) are internalized and turned toward the individual (Corrigan & Bink, 2016). Label avoidance is an additional social construct in which individuals avoid being associated with mental health labels such as being seen purchasing medication and/or going into a therapy clinic, in order to avoid being viewed in a negative or discriminatory fashion. Structural stigma is present when individuals with mental illness are intentionally targeted by private and

government organization where policies enacted limit their opportunities (Corrigan et al., 2014). Some participants may not realize the nuances of mental health stigma or that they may be internalizing self-stigma or label avoidance stigma due to the continued pervasive external negative beliefs and attitudes surrounding mental health care and illness.

The need to highlight mental health stigma is warranted based on the continued stigmatization of mental health care within the US. One fourth of the participants reported struggles with stigma, whether or not they realized stigma was the underlying root of their barrier. The experiential reports of participants discussed their reality and how stigma affected them through social, self, and label avoidance stigmas. One might argue that this demographic is also facing structural stigma based on the extraordinary cost of the CFTR gene modulating medications, thus making them inaccessible based on the monopoly of Vertex Pharmaceuticals.

Application to Theory

The Role of The Family Systems Genetic Illness Model in the Study

With six in ten individuals facing a chronic physical illness in the US (CDC, 2019), it would be pertinent for all counseling and therapy graduate programs to require (or highly encourage) a course in medical family therapy that utilizes a biopsychosocial model to integrate the entire family unit in the course of treatment planning, psychoeducation, and offering of resources. It is known that annually, there are over 40 million diagnoses of depression and 17 million anxiety diagnoses within the US (NIMH, 2019). It is also known that rates of anxiety and depression have been found to be two to three times higher in the CF community, including caregivers, compared to community

samples according to the TIDES study (Quittner et al., 2014). The current study confirms the struggles with increased rates of anxiety and depression and lower rates of quality of life for caregivers raising a child with CF. Within this study, 45% of participants had a current anxiety diagnosis, over 28% had a depression diagnosis, 6% had a diagnosis of "other" including MDD, PTSD, or personality disorder, almost 25% were currently utilizing psychotropic medication to help with their mental health. Based on the participant scores on the GAD-7 and PHQ-9, it appears that 57% were experiencing elevated symptoms of anxiety, 48% were experiencing elevated symptoms of depression representing potentially undiagnosed anxiety and depression in 12% to 20% of the participants, respectively. Additionally, over 50% of participants stated they were interested in a mental health check for themselves at each of their child's clinic visits. There were also over 26% of participants interested in more than an annual screening dependent upon the resources available and how the information would be used. This information is important to note, as the emotional toll from caregiving burden is high, these parents and families need support, and their responses indicate they want more mental health resources within their child's CF services.

According to Rolland and Williams (2005), there is a lack of sufficient knowledge and skills in healthcare systems that have tried to integrate biomedical and psychosocial care by health and mental health professionals working with family-related issues. The need for family systems centered programs is prevalent, and also increasing, as many therapists are going out on their own to find specialized trainings for illness, disability, and loss. While it does not make educational or financial sense to mandate education on

all chronic illnesses, a greater emphasis should be placed on how to help families live with the "uninvited guest" of illness and disease.

As discussed in chapter two, the FSGI is a prevention-oriented, resilience-based family-centered model that is integrative, collaborative, systemic, developmental, and takes into consideration changes across time as well as the genetic component to chronic illness and disease (Rolland, 2018). While a traditional medical model tends to be narrowly focused on the diagnosed individual, the FSGI model expands the unit of care to include the family and/or caregiving system and could be a useful guide in training clinicians who are allies in care and working with individuals and families with health-related struggles (Rolland, 2018). It is relevant to note:

> This model is a sharp contrast to the narrow focus on the patient by most current models of intervention in behavioral medicine, consultation-liaison psychiatry, and psychotherapy. At worst, families are relegated to the background; it is recognized that they affect the patient's psychosocial adjustment, but they are not considered to need help with their own stressful challenges. Early intervention acknowledges the importance and concerns of all family members, prevents them from being marginalized, and draws on their potential as vital partners and resources in the treatment process. Further, I view collaboration as a mindful way of resisting pressures to dehumanize the experience of illness and reduce the ill person and a living family system to a diseased patient or disorder (Rolland, 2018, p. 7–8).

From the participant's responses, it was apparent the entire family unit was affected by CF; from parents struggling to find time and resources for care, to juggling the demands of their own personal needs including work, school, mental health, self-care, etc., to balancing the health and wellbeing of other children and family members who may also reside in the household. The FSGI model places a high emphasis on the family as a caregiving unit and team, and recognizes that stressful events, including chronic illness diagnosis and episodes, will have a ripple effect on all relationships; thus it is a vital resource with the potential to foster adaptation and resilience (Rolland, 2018).

Just as ambiguous loss focuses on creating meaning of one's situation, the FSGI model also highlights the importance of the family beliefs systems, how they make meaning of the health conditions faced and how the shared meaning creates an opportunity, as a family, to establish successful coping, adaptation, agency, and mastery (Rolland, 2018). With a future focus for clinical training with a family-centered model of integrative, collaborative, and systemic care, we are given an opportunity to better support the entire family as a whole, and help them navigate the meaning they are making, working through their grief and ambiguous loss, and decide what is most important to them.

Limitations

There are a few limitations to the study to be noted. Some limitations were known at the start of the study and the cost-benefit analysis was determined to move forward despite potential drawbacks. The first limitation is the length of the study. Although participants were aware the time commitment ranged from 35–50 minutes, there was a

significant amount of drop-off in engagement, resulting in almost half of the cases being removed and excluded from the results. Participation dropped from 297 to 155 completing the entire survey. This may have been due to emotional discomfort or distress based on the study's content or simply survey fatigue. Future studies may benefit from scaling down the survey length in order to avoid participant fatigue as well as keeping it open for a longer amount of time in order to obtain a higher number of participants.

The self-report aspect of this study could be a limitation. If participants needed clarification on any of the questions, there was no opportunity for this to occur due to the online asynchronous format. Along with the self-report aspect of the study, an additional limitation is the online format itself and the lack of face-to-face contact in the experiential question section, where much of the nonverbal body language and communication nuances were missed, thus lacking the option to delve deeper into any of the qualitative questions where it may have been needed. In formulating the study, this limitation was known in advance, but retained in order to increase the probability of recruiting a higher number of participants, specifically male caregivers. The current study did not have a control "well population" for community comparison and was excluded based on wanting to explore the between gender differences, specifically of CF caregivers, due to the historical information that female caregivers are generally better represented within CF research (Shardonofsky, Cesario, Fredland, & Landrum, 2019a) and how certain aspects of the CF diagnosis, history, and experience influenced rates of anxiety, depression, and quality of life for the caregiver. In hindsight, a community sample for comparison purposes may have proven beneficial for this study, especially taking into consideration

the influence of the pandemic and the influence it had on participant's emotional well-being.

Implications

The Need for Integrative, Systemic, and Family-Centered Care

The findings from this study support the need to move from an individual-based treatment plan to that of a collaborative, integrative, and systemic family-centered approach. A majority of participants identified day-to-day stressors that much of the well population also experiences including work performance and balance, home struggles such as marital conflict and divorce, parenting differences, other illnesses and diagnoses, juggling multiple family member's schedules, loss of former self into the role of motherhood, fostering children, legal woes, addiction, and existential dread. Parents and caregivers raising children with CF also struggled with finding time for their child's treatments, exhaustion and overwhelm related to keeping their child physically and mentally healthy in the face of a respiratory pandemic, as well as financial concerns to support their life saving medications and medical needs. Participant's ability to process through their struggles may seem exponentially higher to them personally due to their already strenuous daily concerns of the health and wellness of the child diagnosed with CF, as well as caregiving needs of additional non-CF children, partners, and family members. Taking these factors into consideration and the genetic component of cystic fibrosis, incorporating the FSGI Model as a framework of CF services would be pertinent to best serve the needs of this population.

Looking Deeper at Biopsychosocial Factors: Is there a Need?

This study sought to address additional factors that have been lacking in CF research including increased father involvement, CF severity and CFTR gene mutation, and access to CF care clinics and how they influence caregiver mental health. Father participation was lower than expected but had representation similar to previous studies (Shardonofsky, Cesario, Fredland, & Landrum, 2019a); to my knowledge this is the only study of this format conducted without direct access to a CF care clinic to utilize for recruitment assistance. As CF severity is generally determined by FEV_1 and weight, a majority of quantitative studies use this data to quantify severity, with mixed results on impact to caregiver emotional health (Besier & Goldbeck, 2011; Driscoll et al., 2009). This study found no correlation to CFTR genetic mutation and caregiver mental health, and it is uncertain if there are currently any such studies with evidence to support the impact. CF care clinic distance and access also did not influence caregiver anxiety, depression, or quality of life; again, additional research looking into this relationship may be of interest in the future.

Two main goals of this study were to determine the differences in mother and father rates of anxiety, depression, and quality of life, and how the age and sex of their child influences those rates as well. While statistical significance was lacking in parametric testing, likely due to the small sample size of male participants, this study showed fathers experienced more anxious than depressive symptoms. The mothers in the study, as a whole, self-reported high rates of anxiety and depression and lower rates of quality of life. The most interesting finding was that caregivers raising adolescent

females ages 14 and older reported the lowest levels of anxiety and depression and the highest levels of quality of life compared to all other age groups. This study identified caregivers raising children in the birth to six age group as experiencing higher levels of anxiety and depression, but it did not affect caregiver's quality of life. This study's findings contradict previous studies showing that child age and gender have no association on parental rates of anxiety and depression (Cronly et al., 2019). However, mothers raising younger children experienced elevated rates of anxiety within the TIDES study as well (Quittner et al., 2014). A wider range of factors must continue to be investigated to better understand their impact to the emotional health and well-being not only of the diagnosed individual, but the family as a whole.

The Importance of Experiential Themes and Barriers to Care

Incorporating an experiential qualitative component to enhance and inform the numerical, quantitative data highlighted the lived experiences of our participant caregivers and identified five main themes: (1) The CF Priority, (2) Resources and Support, (3) Pandemic Amplifications of Distress, (4) Natural Skepticism and Cautious Optimism, and (5) Barriers to Seeking and Receiving Mental Health Services. Understanding participant experiences of raising a child with CF in the midst of a global pandemic helped to provide insight into not only the challenges and obstacles this group faces but also the adaptation, hope, and resilience that fuels their motivation and determination to continue seeking a cure for CF. Participants highlighted how the struggle to find balance within participant's daily lives and the financial and time intensive burden of caring for a child with CF took an emotional toll. Although CF and

the care and treatment of the diagnosed child is an overarching theme throughout the study, participants did not view this as a specific hindrance to seeking and receiving mental health services. However, the treatment burden and CF coming first led many putting themselves and their emotional health on the backburner, which led to an increased understanding of the barriers this population faces to seeking and receiving mental health services for themselves.

The CF community is working diligently not only to find a cure for CF but has also recognized the psychological impact of the disease, not only on the individual but the caregivers as well, and have worked to have high quality innovative, integrative, and collaborative care within their clinic care centers (CFF, 2019b). While having the numerical data that identifies elevated rates of anxiety and depression within this population and taking steps to implement mental health more into the clinic experience, the next steps are to continue the discussion and work to identify what specifically contributes to these elevated rates of anxiety and depression in caregivers (and patients) and how to best help this population with emotional wellness whilst awaiting a cure. This current study found that CF takes priority in the daily lives of participants. This group lacks resources and support and while they attempt to be optimistic, there is a skepticism buzzing below the surface as they navigate a global pandemic. It has been documented that CF is one of the most chronically burdensome diseases to manage and it is time to address the barriers of this population.

Enhancing the Approach to Mental Health Care

The findings of this study support previous research (Cronly et al., 2019; Quittner et al., 2014) regarding annual mental health screenings for parents raising children with CF while recommending the option of quarterly or biannual screenings based on participant input. It has also recently been documented that an integrated family centered approach be implemented within CF services providing support, resources, and mental health referrals for parents (Besier & Goldbeck, 2011; Cronly et al., 2019) and the FSGI would be a logical and efficient model to incorporate into the CF care clinics.

When considering the current plan for implementing mental health care services within CF clinics, it appears to be short-sighted and potentially falling short of the trajectory of advancement within the community. There are amazing strides being made to decrease physical symptoms and increase not only quality of life, but the life expectancy of diagnosed individuals. While the emotional well-being and mental health needs of the diagnosed individual are finally being addressed and annual psychological screenings are being offered in select clinics, the current option leaves out 364 days of the year where clients (and their families) may have a mental health concern or crisis that goes unnoticed and/or untreated.

Within this study, participants reported they want increased access, services, and mental health check-ins for themselves and their children; not just annually, but with the option at each quarterly CF clinic visit. A majority 56.6% of parents reported they were interested in enhanced services for themselves, with an additional 26.3% saying they would be interested depending on the resources provided, accessibility and ease of

working with a mental health care professional, and how the information would be used. Parents also reported at a rate of 68.6% they would want the same mental health check-up options for their children, with 22% stating similar concerns of resources, accessibility, and use of the information. With only 17.1% reporting they were not interested in this service for themselves, and 9.3% not interested in additional check-ins for their children, it is important to take the needs of the majority into consideration.

It appears as if the CF community is at the precipice of a shift, where this disease is no longer as debilitating, life shortening, or even considered to be fatal. Action must be taken now while taking in to consideration the advances being made, to adequately plan for the emotional health needs of the diagnosed individual and their families. Proactive planning now may be used to guide CF families throughout the life span as the trajectory of cystic fibrosis continues to change. Looking to the future there is an opportunity now to begin implementing a full scale, one-stop shop for services that are truly systemic, holistic, and take the health needs of the CF diagnosed individual and their families into account.

With this population already struggling to find the time, energy, and resources with coordinating care for their CF child with doctors, their CF care team, and all the other daily items that need to be accounted for and juggled, making time to seek out individualized mental health care falls to wayside, and lack of accessibility to therapists is not helping the situation. It is vital to provide resources targeted specifically to this population that do not contribute to unnecessary obstacles as caretakers are already overwhelmed as a whole. Adding more mental health clinicians into CF clinic services is

crucial in providing ease of access, knowledge, availability, and scheduling for this group.

The CFF prides themselves in providing the highest quality, comprehensive care at accredited centers with a multidisciplinary team of medical professionals to ensure efficiency within the quarterly clinic process (CFF, 2021c). The current required multidisciplinary team consists of a physician, nurse, dietician, respiratory therapist, program coordinator, and social worker; with a research coordinator, pharmacist, physical therapist, and psychologist as recommended care team members (CFF, 2021d). The role of the CF social worker is to help the diagnosed individual by addressing the emotional impact on aspects of living with CF (CFF, 2021d). They may also refer the diagnosed individual out for case management and additional guidance to Compass, a free service for those living with CF. The CF social worker may also refer the patient to a psychologist for additional support during clinic visits or hospitalizations. The CF psychologist may also refer the patient to other therapists or specialists for continued support (CFF, 2021d).

In one clinic visit, the patient is the only person being addressed and may receive three referrals. This process is problematic and inefficient and a perceived barrier to seeking and receiving mental health services. In order for the CF clinics to truly call themselves a multidisciplinary practice, providing access to an in-house CF care partner focused on mental health is vital. Removing the hoops of referral after referral would eliminate the overwhelm of beginning psychotherapeutic treatment. Some may also argue a move from a multidisciplinary practice to an interdisciplinary practice would also be of

the greatest benefit to the diagnosed individual and their families. The CF Care Centers have been recognized as a model of efficiency and effectiveness within chronic disease by the National Institutes of Health (CFF, 2021c), and it is time to add excellence in holistic, systemic, integrated care to this honor by incorporating greater access to mental health professionals into the practice. By adding psychotherapists to the CF care team, there is an opportunity to better meet the needs of the whole family from the physical health of the diagnosed individual to the emotional and mental health of the whole family.

As efforts to minimize the stigma surrounding mental health care continue, and the CFF is working to incorporate an increased presence of preventative mental wellness initiatives within their clinics for the patients and caregivers, it may be wise to consider the input of the participants regarding mental health check-ins for patients and their families. As a majority of CF patients go to routine clinic visits four times a year, this provides an opportunity to gain insight into the mental health needs of the patient as well as their parent caregivers. The PHQ-9 and GAD-7 assessments utilized in this research are readily available online, at no cost, in multiple languages, and take less than five minutes to complete together. They also take into consideration the factors contributing to anxiety and depression over the last 2 weeks with cutoff scores to indicate additional follow-up with a professional may be advisable. While the CQOLCF is not currently available online without permission from the author, it is my belief that should this become a standard practice within the CF clinic visits, requests to release the rights in order to enhance the lives of those within the CF community would be granted. The

CQOLCF takes less than 10 minutes to complete and takes into consideration factors contributing to the perceived quality of life over the past seven days. These assessments could be sent electronically to clients before their child's clinic visit; this would be an opportunity for caregivers to explore additional mental health options for themselves and children, if needed. It would also be nice to see the CF care clinics expand their inclusion from psychologists, social workers, and psychiatrists, to also include licensed professional counselors and marriage and family therapists.

Therapist Accessibility and CF Specific Training

This study appears to be the first of its kind within the US, taking place outside of a CF care center, and looking at both the numerical and experiential data simultaneously, which will help to not only begin to bridge the gap in literature, but also encourage further mixed methods research on a larger scale. It is vital that future mental health professionals have the opportunity to learn more about family-centered, systemic medical models of family therapy care, and that current clinicians are able to find resources to better support those who experience chronic illness as it impacts so many individuals annually within the US.

While it is important to note therapists should not practice outside their scope of knowledge, it is difficult to draw a hard line and determine that all therapists should be knowledgeable about CF, considering the CF population within the US is roughly 30,000, with an approximate total population of 330 million. It is my hope that this study helped to highlight some of the struggles and frustrations these caregivers experienced in order

to minimize the number of barriers they face in seeking and receiving their own much deserved mental health care.

As one participant noted, she is so excited for the future of CF, especially with the introduction of physical health improvements via CFTR gene modulating medications, that she no longer considers CF to be fatal. With the continued advances of the CFF in searching for a cure, this disease will hopefully soon be one where the predicted life expectancy is within the normal range of the general population. Having a better understanding numerically, experientially, and longitudinally of how CF influences the diagnosed individual as well as their family's emotional well-being will help clinicians obtain a better understanding on how to help facilitate interventions that meet their psychological needs. Although much of this study was not statistically significant, the experiential input is priceless in helping to move the needle forward and providing mental health clinicians a foundation of knowledge to work more effectively with the CF community from a family-centered, resilience-based approach, because these families need and deserve mental health care that is respectful and responsive to their specific chronic illness circumstances.

Suggestions for Future Research

Although a turnout of almost 10% of the participants being represented by male caregivers was encouraging, there is a continued need to have increased participation by fathers in order to give their experiences a voice. Future studies with a similar distribution of male and female caregiver participants could be beneficial in better understanding the differences in their experiences. Additionally, attempts to determine

how CF severity and genetic mutation influence emotional health and quality of life was rejected due to the lack of participants raising children with rarer gene mutations and the majority of the population having children with mild CF. Future studies may benefit from having a larger, heterogeneous sample with a more diverse range of participant genetic mutations represented. It would also be helpful to focus on resilience, adaptation, and coping in order to understand how individuals with children with moderate and severe CF go through the process throughout the life cycle.

Many participants discussed their struggles in balancing CF treatments into their daily schedules while also ensuring their other children were receiving adequate care, attention, and affection. Some identified feeling overwhelmed when being torn between the CF routine and tending to the needs of their other children. Those raising toddlers expressed difficulty in managing the needs and care of both children. While the focus of this specific study was on the emotional health of caregivers raising children with CF, it would be beneficial to also study the impact on siblings of children with CF and how other children within the household are also affected. As CF is a lifelong illness that impacts not only the diagnosed individual, but the entire family, it will be important to also address the emotional health concerns of CF siblings moving forward.

Finally, this study sought to determine how the child's sex and gender influenced emotional well-being. In a future study, having a better representation of parents raising children of both sexes in all three age groups to help provide additional clarification on this topic would be beneficial. Again, focusing on strengths, coping, adapting, and resilience and how parents go through the life cycle with sons versus daughters. Future

studies may also benefit from conducting smaller groups comparing adult emotional wellness within the same household to better identify and understand the differences between caregiving experiences. In hindsight, it may have been important to note where the CF diagnosed child fell in the birth order of the number of children of participants. Additionally, the emotional impact of raising a son versus a daughter in the youngest age group could also be explained by the size of the sample; of the participants, 64.2% were reporting raising a son versus 35.8% raising a daughter. Nevertheless, this could potentially be an area where parents need more support and more resources from all areas as they navigate the early years of raising a child, as well as navigating the world of CF.

Conclusions

Overall, this study looked at caregiver rates of anxiety, depression, and quality of life and the psychosocial factors encompassed by raising a child with a cystic fibrosis diagnosis. This study expanded on previous quantitative studies which highlighted the elevated rates of anxiety and depression and lower levels of quality of life of caregivers raising a child with CF and incorporated a qualitative experiential piece to help understand the true lived experiences of this population.

The results of this study found that caregivers raising children from birth to six struggled most with anxiety and depression when compared to the older age brackets of children of both sexes. Caregivers raising adolescent daughters ages 14 and older reported the lowest levels of anxiety and depression and the highest rates of quality of life. Distance to CF clinics did not have an effect on parental emotional well-being, nor did the child's CF severity or genetic mutation. Fathers struggled more with symptoms of

anxiety than depression. Over half of the study's mothers reported elevated rates of both anxiety and depression. While there was a lack of statistical significance between male and female caregiver rates of anxiety, depression, and quality of life, mothers continue to report elevated rates of anxiety and depression and a lower quality of life than compared to fathers; this could be explained by the distribution discrepancy and the overwhelming majority of participants being represented by mothers.

From the experiential component of the study, the accounts provided found overwhelmingly that the influence of CF takes the top priority within the family unit. Participants struggled with resources such as time, finances, and navigating insurance. Many reported a disconnected lack in social and familial support. There was natural skepticism present throughout responses from views on CFTR modulating medication, the coronavirus, and seeking mental health services. While the skepticism may have been present, it appeared that many also held a cautious optimism that helped them to create meaning, shift their perspective and adapt, and cope by seeking joy and happiness in the present moment, despite their circumstances. Insight into participant's barriers to seeking and receiving mental health services also provided ample opportunity to acknowledge the role of mental health stigma within the process and how access to mental health clinicians who are knowledgeable with chronic illness, especially CF is lacking. Participant responses help us to recognize this group requires additional resources, support, and an active plan to dismantle the obstacles they face in putting themselves and their emotional wellbeing first.

Through their participation, parents and caregivers helped to bridge the gap between understanding the high numerical data reflecting elevated rates of anxiety, depression, and lower rates of quality of life with an account of their daily experiences which also contribute to barriers in seeking and receiving individual mental health care. As medical advances continue for CF patients and life expectancy is increasing, it is vital to understand how to best serve this population with mental health care concerns throughout the life span. Therefore, it is imperative that continued efforts are made to research and understand the connection between the numerical data and the influence on parental emotional health. There is also an ongoing need to address the barriers to seeking and receiving mental health care when raising a chronically ill child with CF. Finally, the dialogue must continue in identifying the best practices of treatment and psychological intervention within the mental health community, inside and outside of the CF clinics.

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APPENDIX A

Facebook Group Posting Authorizations

TWU

Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu>

Online Consent Authorization Request

2 messages

Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu> Thu, Apr 2, 2020 at 3:19 PM
To: carolyehgarner@gmail.com

Good Afternoon Carol!

I hope this email finds you well. Thank you for providing verbal interest and consent to share a request for research participants in my dissertation study, "An Exploration of the Experiences of Caregivers Raising Children with a Cystic Fibrosis Diagnosis and the Relationship between rates of Anxiety, Depression, and Quality of Life: An Online Exploratory Mixed-Methods Study" within your online Facebook group, "The Distillery."

If you could respond to this email expressing written consent to share within your group, that I may include in my dissertation proposal appendices, I would greatly appreciate it.

Please be advised you are under no obligation to share my request, are under no pressure to encourage anyone to participate, and may revoke permission at any time.

If this is agreeable to you, please simply respond to this email with the following, "I, Carol Yeh-Garner, CEO & President of A Well Lived Life, Inc & Founder of The Distillery, acknowledge my consent and authority to share a request for dissertation research participants in my online Facebook groups."

Thank you for supporting my research endeavors. If you have any questions or concerns please reach out at any time.

In Gratitude,

Kristina Dingus Keuhlen, MA, MS, LMFT Associate
Texas Woman's University Marriage and Family Therapy Doctoral Candidate

---

Carol Yeh-Garner <carolyehgarner@gmail.com> Fri, Apr 3, 2020 at 2:00 PM
To: Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu>

Hi-

I, Carol Yeh-Garner, CEO & President of A Well Lived Life, Inc & Founder of The Distillery, acknowledge my consent and authority to share a request for dissertation research participants in my online Facebook groups.

Live, Laugh, Love…Oil On,

Carol Yeh-Garner
Young Living Royal Crown Diamond Independent Distributor #1533467
858-837-1259
www.carolyehgarner.com

[Quoted text hidden]

APPENDIX B

Social Media Recruitment Flyer

Are you a caregiver raising a child with Cystic Fibrosis?

Research indicates caregivers of chronically ill children experience higher rates of anxiety and depression and lower rates of quality of life. Within the CF community these rates have been found to be 2-3 times higher than the general population. I am interested in understanding what is contributing to increased rates of emotional health struggles for CF caregivers.

Historically, female caregivers are better represented in CF research, therefore male caregivers are especially encouraged to participate.

My study is titled: "An Exploration of the Experiences of Caregivers Raising Children with a Cystic Fibrosis Diagnosis and the Relationship Between Rates of Anxiety, Depression, and Quality of Life: An Online Exploratory Mixed-Methods Study."

For my dissertation research as a marriage and family therapy doctoral student, I am interested in exploring the lived experiences of caregivers raising children with a cystic fibrosis diagnosis and the numerical connection to anxiety, depression, and quality of life. Your participation could contribute to how therapists are prepared to work with individuals and families within the CF community. If you would like to share your story and participate, please read below and contact us.

This study is completely online. It is a series of assessments to identify numerical input, and open-ended questions to incorporate participant perspectives and experiences. The study time commitment is approximately 40-50 minutes. Participation is voluntary and you may withdrawal at any time.

The greatest risks of this study include potential loss of confidentiality, loss of time, fatigue, emotional discomfort and/or emotional distress.

If you are interested, have questions, or would like to participate, please contact me, Kristina Dingus, at 972-841-9171 or email DingusResearch@gmail.com. You may also contact my research advisor, Linda Ladd, PhD, PsyD at 940-391-0834 or Lladd@twu.edu.

There is a potential risk of loss of confidentiality in all email, downloading, electronic meetings, and internet transactions.

APPENDIX C

Caregiver Demographic Questionnaire

Caregiver Demographic Questionnaire

Directions: Please complete the following, to the best of your ability, with as accurate information as possible. You will be asked questions regarding your age range, race/ethnicity, marital and employment status, annual income, education, and mental health.

Please check this box if you are aware of your child's other parent caregiver completing this study:

Yes
No

The following 22 questions have to do with your (parent/caregiver) information.

1. Please select the response that best describes your relationship with the child:
    a. Mother/Stepmother/Foster mother/Adoptive mother
    b. Father/Stepfather/Foster father/Adoptive father
    c. Grandparent
    d. Aunt/Uncle
    e. Sibling
    f. Legal Guardian
    g. Other
2. Please select your gender:
    a. Male
    b. Female
    c. Other, please identify:
3. Please identify your age at your last birthday:
    a. 21-29
    b. 30-39
    c. 40-49
    d. 50-59
    e. 60-69
    f. 70+
4. Please identify your current relationship status:
    a. Single (Never Married)
    b. Married, or in a domestic partnership
    c. Widowed
    d. Separated
    e. Divorced
5. Are you of Hispanic, Latino, or of Spanish origin?
    a. Yes
    b. No
6. How would you describe yourself?
    a. American Indian or Alaska Native
    b. Asian

c. Black or African American
d. Native Hawaiian or Other Pacific Islander
e. White
f. Other

7. Please identify your current religious beliefs, if any (listed in alphabetical order):
    a. Agnostic (not sure if there is a God)
    b. Anglican
    c. Atheist (do not believe in God)
    d. Baha'i
    e. Baptist
    f. Buddhist
    g. Catholic
    h. Christian
    i. Evangelical
    j. Hindu
    k. Jehovah's Witness
    l. Jewish
    m. Lutheran
    n. Methodist
    o. Mormon
    p. Muslim
    q. Nothing in particular
    r. Orthodox
    s. Other
    t. Prefer not to state
    u. Protestant
    v. Sikh
    w. Spiritual, but do not consider myself religious
    x. Unitarian
8. Please identify the highest educational level of education completed:
    a. Less than a high school diploma
    b. High School Diploma/GED
    c. Some college/ no degree
    d. Trade School Certification
    e. Associate degree (e.g. AA, AS)
    f. Bachelor degree (e.g. BA, BS)
    g. Master's degree (e.g. MA, MS, MEd)
    h. Professional degree (e.g. MD, DDS, DVM)
    i. Doctoral degree (e.g. PhD, PsyD, EdD)
    j. Other (please specify) _____
9. Please identify your gross annual household income:
    a. Below $25,000
    b. $26,000-$40,000
    c. $40,001-$60,000

d. $60,001-$80,000
e. $80,001-$100,000
f. $100,001-$150,000
g. Above $150,001

10. Please identify your employment status:
    a. Employed Full-time (40 or more hours per week)
    b. Employed Part-time (up to 39 hours per week)
    c. Unemployed and currently looking for work
    d. Unemployed and not currently looking for work
    e. Temporary/Contract
    f. Student
    g. Stay-at-Home Caregiver
    h. Retired
    i. Unable to work
11. Please identify the number and age of total children who live in your home at least 50% of the time:
    a. Open Response Format
12. Please identify your insurance coverage:
    a. Yes
    b. If yes, please indicate type: through employer, private, through CFF
    c. No
13. Please identify if you have ever been diagnosed with anxiety:
    a. Yes
    b. No
14. Please identify if you are currently diagnosed with anxiety:
    a. Yes
    b. No
15. Please identify if you have ever been diagnosed with depression:
    a. Yes
    b. No
16. Please identify if you are currently diagnosed with depression:
    a. Yes
    b. No
17. Please identify if you have ever been diagnosed with another mental health illness/disorder:
    a. Yes, please specify: _____
    b. No
18. Please identify if you are currently diagnosed with another mental health illness/disorder:
    a. Yes, please specify: _____
    b. No
19. Please identify if you are currently under the care of a mental health professional (therapist, counselor, psychologist, psychiatrist):
    a. Yes

    b. No
20. Please identify if you have ever been under the care of a mental health professional (therapist, counselor, psychologist, psychiatrist):
    a. Yes
    b. No
21. Please identify if you are currently taking any psychotropic medication to treat diagnosed mental health illness/disorder:
    a. Yes
    b. No
22. Please identify if you have ever taken any psychotropic medication to treat diagnosed mental health illness/disorder:
    a. Yes
    b. No

APPENDIX D

Child Demographic Questionnaire

As reported by the participant caregiver

Child Demographic Questionnaire

Directions: Please answer the following questions in this questionnaire for your child with a cystic fibrosis diagnosis unless otherwise indicated. For consistency purposes, please identify a referent (the same child) child for your responses. You will be asked about your child's education, physical health characteristics including CFTR gene mutation, FEV_1/ commonly referred to as pulmonary function test (PFT), BMI, hospitalizations, and mental health.

1. Please identify the number and age of total children who live in your home at least 50% of the time:
    a. Open Response Format
2. Please identify the number and age of children diagnosed with cystic fibrosis:
    a. Open Response Format
3. Please identify your child's type of education:
    a. Public school system
    b. Private school
    c. Montessori
    d. Homeschool
    e. Other (please specify): _____
4. Please identify the sex, current age, and age of diagnosis of your child:
    a. M/F Current Age: Age of Diagnosis:
    b. M/F Current Age: Age of Diagnosis:
    c. M/F Current Age: Age of Diagnosis:
    d. M/F Current Age: Age of Diagnosis:
5. Please identify the CFTR gene mutation for your child:
    a. Child 1:
    b. Child 2:
    c. Child 3:
    d. Child 4:
    e. Don't Know
6. Please identify the most recent FEV_1/PFT recorded for your child:
    a. Child 1:
    b. Child 2:
    c. Child 3:
    d. Child 4:
    e. Don't Know
7. Please identify the most recent BMI recorded for your child:
    a. Child 1:
    b. Child 2:
    c. Child 3:
    d. Child 4:
    e. Don't Know

8. Please identify the most recent weight recorded for your child:
    a. Child 1:
    b. Child 2:
    c. Child 3:
    d. Child 4:
    e. Don't Know
9. Please identify the severity of CF for your child as diagnosed by their medical doctor:
    a. Child 1:
    b. Child 2:
    c. Child 3:
    d. Child 4:
    e. Unsure:
10. Have any of your children ever been on the lung/organ transplant list?
    a. Yes
    b. No
    c. If yes, please identify which number child:
11. Have any of your children previously received a lung/organ transplant?
    a. Yes
    b. No
    c. If yes, please identify which number child:
12. Has your child been hospitalized for a tune-up within the past six months?
    a. Yes
    b. No
    c. If yes, please identify which number child:
13. Has your child required nasal surgery within the past six months?
    a. Yes
    b. No
    c. If yes, please identify which number child:
14. Has your child ever had a positive culture for any of the following bacterium:
    a. Y/N: *Pseudomonas aeruginosa*
        i. If yes, please identify which number child:
    b. Y/N: *Mycobacterium abscessus*
        i. If yes, please identify which number child:
    c. Y/N: *Burkholderia cenopacia*
        i. If yes, please identify which number child:
15. Has your child been diagnosed with any additional illness(es) as a result of their cystic fibrosis diagnosis?
    a. Yes
    b. If yes, please specify:
        i. Cystic Fibrosis Related Diabetes (CFRD)
        ii. Cystic Fibrosis Related Metabolic Syndrome (CRMS)
        iii. Cystic Fibrosis Related Asthma
        iv. Cystic Fibrosis Related Bone Disease

v. Cystic Fibrosis Related Liver Disease
vi. Other, please identify: _____

c. No

16. Please identify your child's CF Clinic:
    a. The online survey will list all 130 locations in a drop down menu that participants may select.
17. On average, how much time do you spend each day on your child's treatments, administering medication, planning nutrition, and sanitizing?
    a. Open Format Response
18. Please identify if your child is currently diagnosed with anxiety:
    a. Yes
    b. No
19. Please identify if your child has ever been diagnosed with anxiety:
    a. Yes
    b. No

20. Please identify if your child currently diagnosed with depression:
    a. Yes
    b. No
21. Please identify if your child has ever been diagnosed with depression:
    a. Yes
    b. No
22. Please identify if your child is currently diagnosed with another mental health illness/disorder:
    a. Yes, please specify: _____
    b. No
23. Please identify if your child has ever been diagnosed with another mental health illness/disorder:
    a. Yes, please specify: _____
    b. No
24. Please identify if your child is currently under the care of a mental health professional (therapist, counselor, psychologist, psychiatrist) who is not your CF social worker:
    a. Yes
    b. No
25. Please identify if your child has ever been under the care of a mental health professional (therapist, counselor, psychologist, psychiatrist) who is not your CF social worker:
    a. Yes
    b. No
26. Please identify if your child is currently taking any psychotropic medication to treat diagnosed mental health illness/disorder:
    a. Yes
    b. No

27. Please identify if your child has ever taken any psychotropic medication to treat diagnosed mental health illness/disorder:
    a. Yes
    b. No

APPENDIX E

General Anxiety Disorder-7 (GAD-7)

GAD-7

Over the last 2 weeks, how often have you been bothered by the following problems? *(Use "✔" to indicate your answer)*	Not at all	Several days	More than half the days	Nearly every day
1. Feeling nervous, anxious or on edge	0	1	2	3
2. Not being able to stop or control worrying	0	1	2	3
3. Worrying too much about different things	0	1	2	3
4. Trouble relaxing	0	1	2	3
5. Being so restless that it is hard to sit still	0	1	2	3
6. Becoming easily annoyed or irritable	0	1	2	3
7. Feeling afraid as if something awful might happen	0	1	2	3

(***For office coding: Total Score T____=_____+_____+_____+_____***)

Developed by Drs. Robert L. Spitzer, Janet B.W. Williams, Kurt Kroenke and colleagues, with an educational grant from Pfizer Inc. No permission required to reproduce, translate, display or distribute.

APPENDIX F

Patient Health Questionnaire-9 (PHQ-9)

PATIENT HEALTH QUESTIONNAIRE-9 (PHQ-9)

Over the last 2 weeks, how often have you been bothered by any of the following problems? *(Use "✔" to indicate your answer)*	**Not at all**	**Several days**	**More than half the days**	**Nearly every day**
1. Little interest or pleasure in doing things	0	1	2	3
2. Feeling down, depressed, or hopeless	0	1	2	3
3. Trouble falling or staying asleep, or sleeping too much	0	1	2	3
4. Feeling tired or having little energy	0	1	2	3
5. Poor appetite or overeating	0	1	2	3
6. Feeling bad about yourself — or that you are a failure or have let yourself or your family down	0	1	2	3
7. Trouble concentrating on things, such as reading the newspaper or watching television	0	1	2	3
8. Moving or speaking so slowly that other people could have noticed? Or the opposite — being so fidgety or restless that you have been moving around a lot more than usual	0	1	2	3
9. Thoughts that you would be better off dead or of hurting yourself in some way	0	1	2	3

FOR OFFICE CODING 0 + _____ + _____ + _____

=Total Score: _____

If you checked off any problems, how difficult have these problems made it for you to do your work, take care of things at home, or get along with other people?

Not difficult at all	**Somewhat difficult**	**Very difficult**	**Extremely difficult**
○	○	○	○

Developed by Drs. Robert L. Spitzer, Janet B.W. Williams, Kurt Kroenke and colleagues, with an educational grant from Pfizer Inc. No permission required to reproduce, translate, display or distribute.

APPENDIX G

Caregiver Quality of Life Cystic Fibrosis (CQOLCF) Scale

CAREGIVER QUALITY OF LIFE CYSTIC FIBROSIS (CQOLCF) SCALE©

INSTRUCTIONS: Below is a list of statements about the role of caring for someone with Cystic Fibrosis. By specifying one number per line, please show how true each statement has been for you during the past 7 days.

RESPONSE OPTIONS:

0 = Not at all 1 = A little bit 2 = Somewhat 3 = Quite a bit 4 = Very much

During the Past 7 Days:

_____(1) It bothers me that my daily routine is changed.

_____(2) My sleep is less restful.

_____(3) The need to protect my loved one bothers me.

_____(4) I am satisfied with my sex life.

_____(5) It is a challenge to maintain my outside interests.

_____(6) I am satisfied with the support I get from my family.

_____(7) I feel frustrated.

_____(8) My economic future is uncertain.

_____(9) I fear my loved one will die.

_____(10) I am discouraged about the future.

_____(11) My level of stress and worry has increased.

_____(12) My sense of spirituality has increased.

_____(13) It bothers me, limiting my focus day-to-day.

_____(14) I feel sad.

_____(15) I feel under increased mental strain.

_____(16) I get support from my friends and neighbors.

_____(17) Family communication has increased.

_____(18) I am concerned about our insurance coverage.

_____(19) I feel nervous.

_____(20) I worry about the impact my loved one's illness has had on my other children or other family members.

_____(21) I have difficulty dealing with my loved one's changing eating habits.

CAREGIVER QUALITY OF LIFE CYSTIC FIBROSIS (CQOLCF) SCALE
(Continued)

RESPONSE OPTIONS:

0 = Not at all 1 = A little bit 2 = Somewhat 3 = Quite a bit 4 = Very much

During the Past 7 Days:

_____(22) I have developed a closer relationship with my loved one.

_____(23) I feel adequately informed about my loved one's illness.

_____(24) It bothers me that I need to be available to go to so many of my loved one's appointments.

_____(25) I fear the adverse effects of treatment on my loved one.

_____(26) The responsibility I have for my loved one's care at home is overwhelming.

_____(27) I am glad that my focus is on keeping my loved one well.

_____(28) I feel guilty.

_____(29) It bothers me that my priorities have changed.

_____(30) My daily life is imposed upon.

_____(31) It upsets me to see my loved one deteriorate.

_____(32) The need to manage my loved one's symptoms/illness is overwhelming.

_____(33) I have more of a positive outlook on life since my loved one's diagnosis.

_____(34) I am under a financial strain.

_____(35) It bothers me that other family members have not shown interest in taking care of my loved one.

Suggested Citation: Boling W, Macrina DM, Clancy JP. (2003). The Caregiver Quality of Life Cystic Fibrosis (CQOLCF) Scale: Modification and Validation of a Scale to Measure Quality of Life in the Family Caregivers of Patients with Cystic Fibrosis. Quality of Life Research Journal. 12(8): 1119-1126.

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APPENDIX H

The Cystic Fibrosis Questionnaire Revised (CFQ-R)

CFQ-R Selection Criteria

Please select how the CFQ-R Questionnaire is being implemented:

A. My child completed the questionnaire independently (Children Ages 12 and 13 Self-Report Format; Adolescent and Adults [Patients 14 Years Old and Older])

B. I interviewed my child and recorded their responses (Children Ages 6-11 Interviewer Format; Children Ages 12 to 13 Self-Report Format)

C. I completed the questionnaire on behalf of my child without their input (Parent/Caregivers of Children Ages 6 to 13; Children Ages 12 and 13 Self-Report Format; Adolescents and Adults [Patients 14 Years Old and Older])

D. I have a child between one day old but younger than six and am not completing a CFQ-R, as it is not available.

CFQ-R

Children Ages 6 to 11 (Interviewer Format)

CYSTIC FIBROSIS QUESTIONNAIRE-REVISED

This questionnaire is formatted for use by an interviewer. Please use this format for younger children. For older children who seem able to read and answer the questions on their own, such as 12 and 13 year olds, use this questionnaire in its self-report format.

There are directions for the interviewer for each section of the questionnaire. Directions that you should *read* to the child are indicated by quotation marks. Directions that you are to *follow* are underlined and set in italics.

Interviewer: *Please ask the following questions.*

A. What is your date of birth?

Date ☐☐ ☐☐ ☐☐☐☐
Mo Day Year

B. Are you?
☐ Male ☐ Female

C. During the **past two weeks**, have you been on vacation or out of school for reasons **NOT** related to your health?

☐ Yes ☐ No

D. Which of the following best describes your racial background?
- ☐ Caucasian
- ☐ African American
- ☐ Hispanic
- ☐ Asian/Oriental or Pacific Islander
- ☐ Native American or Native Alaskan
- ☐ Other (please describe)

  _____
- ☐ Prefer not to answer this question

E. What grade are you in now?
(If summer, grade just finished)
- ☐ Kindergarten
- ☐ 1st grade
- ☐ 2nd grade
- ☐ 3rd grade
- ☐ 4th grade
- ☐ 5th grade
- ☐ 6th grade
- ☐ 7th grade
- ☐ Not in school

Interviewer: *Please read the following to the child:*

"These questions are for children like you who have cystic fibrosis. Your answers will help us understand what this disease is like and how your treatments help you. So, answering these questions will help you and others like you in the future."

"For each question that I ask, choose one of the answers on the cards I'm about to show you."

Present the orange card to the child.

"Look at this card and read with me what it says: **very true, mostly true, somewhat true, not at all true**."

"Here's an example: If I asked you if it is **very true, mostly true, somewhat true, not at all true** that elephants can fly, which one of the four answers on the card would you choose?"

Present the blue card to the child.

"Now, look at this card and read with me what it says: **always / often / sometimes / never**."

"Here's another example: If I asked you if you go to the moon **always, often, sometimes, or never**, which answer on the card would you choose?"

Present the orange card to the child.

"Now, I will ask you some questions about your everyday life."

"Tell me if you find the statements I read to you to be **very true, mostly true, somewhat true, or not at all true**."

Please check the box indicating the child's response.

"During the past **two weeks**":	**Very True**	**Mostly True**	**Somewhat True**	**Not at all True**
1. You were able to walk as fast as others	☐	☐	☐	☐
2. You were able to climb stairs as fast as others	☐	☐	☐	☐
3. You were able to run, jump, and climb as you wanted	☐	☐	☐	☐
4. You were able to run as quickly and as long as others	☐	☐	☐	☐
5. You were able to participate in sports that you enjoy (e.g., swimming, soccer, dancing or others)	☐	☐	☐	☐
6. You had difficulty carrying or lifting heavy things such as books, your school bag, or a backpack	☐	☐	☐	☐

Children Ages 6 to 11 (Interviewer Format)

CYSTIC FIBROSIS QUESTIONNAIRE-REVISED

Interviewer: *Present the blue card to the child.*

Please check the box indicating the child's response.

"And during these past **two weeks**, tell me how often":	**Always**	**Often**	**Sometimes**	**Never**
7. You felt tired	☐	☐	☐	☐
8. You felt mad	☐	☐	☐	☐
9. You felt grouchy	☐	☐	☐	☐
10. You felt worried	☐	☐	☐	☐
11. You felt sad	☐	☐	☐	☐
12. You had trouble falling asleep	☐	☐	☐	☐
13. You had bad dreams or nightmares	☐	☐	☐	☐
14. You felt good about yourself	☐	☐	☐	☐
15. You had trouble eating	☐	☐	☐	☐
16. You had to stop fun activities to do your treatments	☐	☐	☐	☐
17. You were pushed to eat	☐	☐	☐	☐

Interviewer: *Present the orange card to the child.*

"Now tell me if you find the statements I read to you to be **very true, mostly true, somewhat true,** or **not at all true**."

Please check the box indicating the child's response.

"During the past **two weeks**":	**Very True**	**Mostly True**	**Somewhat True**	**Not at all True**
18. You were able to do all of your treatments	☐	☐	☐	☐
19. You enjoyed eating	☐	☐	☐	☐
20. You got together with friends a lot	☐	☐	☐	☐
21. You stayed at home more than you wanted to	☐	☐	☐	☐
22. You felt comfortable sleeping away from home (at a friend or family member's house or elsewhere)	☐	☐	☐	☐
23. You felt left out	☐	☐	☐	☐

CFQ-R

Children Ages 6 to 11 (Interviewer Format)

CYSTIC FIBROSIS QUESTIONNAIRE-REVISED

"During the past **two weeks**":	Very True	Mostly True	Somewhat True	Not at all True
24. You often invited friends to your house	☐	☐	☐	☐
25. You were teased by other children	☐	☐	☐	☐
26. You felt comfortable discussing your illness with others (friends, teachers	☐	☐	☐	☐
27. You thought you were too short	☐	☐	☐	☐
28. You thought you were too thin	☐	☐	☐	☐
29. You thought you were physically different from others your age	☐	☐	☐	☐
30. Doing your treatments bothered you	☐	☐	☐	☐

Interviewer: ***Present the blue card to the child again***

Please check the box indicating the child's response.

"Tell me how often in the past **two weeks**":	Always	Often	Sometimes	Never
31. You coughed during the day	☐	☐	☐	☐
32. You woke up during the night because you were coughing	☐	☐	☐	☐
33. You had to cough up mucus	☐	☐	☐	☐
34. You had trouble breathing	☐	☐	☐	☐
35. Your stomach hurt	☐	☐	☐	☐

*Please be sure **all** the questions have been answered.*

THANK YOU FOR YOUR COOPERATION!

Children Ages 12 and 13 (Self-report Format)

CYSTIC FIBROSIS QUESTIONNAIRE-REVISED

These questions are for children like you who have cystic fibrosis. Your answers will help us understand what this disease is like and how your treatments help you. So, answering these questions will help you and others like you in the future.

Please answer all the questions. There are no right or wrong answers! If you are not sure how to answer, choose the response that seems closest to your situation.

Please fill in the answer or check the box that matches your response to these questions.

A. What is your date of birth?

Date ☐☐/☐☐/☐☐☐☐
Mo Day Year

B. Are you?

☐ Male ☐ Female

C. During the **past two weeks**, have you been on vacation or out of school for reasons **NOT** related to your health?

☐ Yes ☐ No

D. Which of the following best describes your racial background?

☐ Caucasian
☐ African American
☐ Hispanic
☐ Asian/Oriental or Pacific Islander
☐ Native American or Native Alaskan
☐ Other (please describe)
_____
☐ Prefer not to answer this question

E. What grade are you in now?

(If summer, grade you just finished)

☐ 5th grade
☐ 6th grade
☐ 7th grade
☐ 8th grade
☐ 9th grade
☐ Not in school

CFQ-R

Children Ages 12 and 13 (Self-report Format)

CYSTIC FIBROSIS QUESTIONNAIRE-REVISED

Please check the box matching your response.

In the past **two weeks**:	Very True	Mostly True	Somewhat True	Not at all True
1. You were able to walk as fast as others	☐	☐	☐	☐
2. You were able to climb stairs as fast as others	☐	☐	☐	☐
3. You were able to run, jump, and climb as you wanted	☐	☐	☐	☐
4. You were able to run as quickly and as long as others	☐	☐	☐	☐
5. You were able to participate in sports that you enjoy (e.g., swimming, soccer, dancing or others)	☐	☐	☐	☐
6. You had difficulty carrying or lifting heavy things such as books, your school bag, or a backpack	☐	☐	☐	☐

Please check the box matching your response.

And during these past **two weeks**, indicate how often:	Always	Often	Sometimes	Never
7. You felt tired	☐	☐	☐	☐
8. You felt mad	☐	☐	☐	☐
9. You felt grouchy	☐	☐	☐	☐
10. You felt worried	☐	☐	☐	☐
11. You felt sad	☐	☐	☐	☐
12. You had trouble falling asleep	☐	☐	☐	☐
13. You had bad dreams or nightmares	☐	☐	☐	☐
14. You felt good about yourself	☐	☐	☐	☐
15. You had trouble eating	☐	☐	☐	☐

Please check the box matching your response.

And during these past **two weeks**, indicate how often:

	Always	Often	Sometimes	Never
16. You had to stop fun activities to do your treatments	☐	☐	☐	☐
17. You were pushed to eat	☐	☐	☐	☐

Please check the box matching your response.

During the past **two weeks**:

	Very True	Mostly True	Somewhat True	Not at all True
18. You were able to do all of your treatments	☐	☐	☐	☐
19. You enjoyed eating	☐	☐	☐	☐
20. You got together with friends a lot	☐	☐	☐	☐
21. You stayed at home more than you wanted to	☐	☐	☐	☐
22. You felt comfortable sleeping away from home (at a friend or family member's house or elsewhere)	☐	☐	☐	☐
23. You felt left out	☐	☐	☐	☐
24. You often invited friends to your house	☐	☐	☐	☐
25. You were teased by other children	☐	☐	☐	☐
26. You felt comfortable discussing your illness with others (friends, teachers)	☐	☐	☐	☐
27. You thought you were too short	☐	☐	☐	☐
28. You thought you were too thin	☐	☐	☐	☐
29. You thought you were physically different from others your age	☐	☐	☐	☐
30. Doing your treatments bothered you	☐	☐	☐	☐

Please check the box matching your response.

Let us know how often in the past **two weeks**:	Always	Often	Sometimes	Never
31. You coughed during the day	☐	☐	☐	☐
32. You woke up during the night because you were coughing	☐	☐	☐	☐
33. You had to cough up mucus	☐	☐	☐	☐
34. You had trouble breathing	☐	☐	☐	☐
35. Your stomach hurt	☐	☐	☐	☐

Please be sure all the questions have been answered.

THANK YOU FOR YOUR COOPERATION!

CFQ - R

Parents/Caregivers (Children Ages 6 to 13)

CYSTIC FIBROSIS QUESTIONNAIRE - REVISED

Understanding the impact of your child's illness and treatments on his or her everyday life can help your healthcare team keep track of your child's health and adjust his or her treatments. For this reason, we have developed a quality of life questionnaire specifically for parents of children with cystic fibrosis. We thank you for your willingness to complete this questionnaire.

Instructions: The following questions are about the current state of your child's health, as he or she perceives it. This information will allow us to better understand how he or she feels in everyday life.
Please answer all the questions. There are no right or wrong answers! If you are not sure how to answer, choose the response that seems closest to your child's situation.

Section I. Demographics

Please fill in the information or check the box indicating your answer.

A. What is your **child's** date of birth?

Date ☐☐ ☐☐ ☐☐☐☐
Mo Day Year

B. What is your relationship to the child?

- ☐ Mother
- ☐ Father
- ☐ Grandmother
- ☐ Grandfather
- ☐ Other relative
- ☐ Foster mother
- ☐ Foster father
- ☐ Other (please describe)

_____

C. Which of the following best describes your child's racial or ethnic background?

- ☐ Caucasian
- ☐ African American
- ☐ Hispanic
- ☐ Asian/Oriental or Pacific Islander
- ☐ Native American or Native Alaskan
- ☐ Other (please describe)

_____

- ☐ Prefer not to answer this question

D. During the **past two weeks**, has your child been on vacation or out of school for reasons **NOT** related to his or her health?

☐ Yes ☐ No

E. What is **your** date of birth?

Date ☐☐ ☐☐ ☐☐☐☐
Mo Day Year

F. What is your current marital status?

- ☐ Single/never married
- ☐ Married
- ☐ Widowed
- ☐ Divorced
- ☐ Separated
- ☐ Remarried
- ☐ With a partner

G. What is the highest grade in school you have completed?

- ☐ Some high school or less
- ☐ High school diploma/GED
- ☐ Vocational school
- ☐ Some college
- ☐ College degree
- ☐ Professional or graduate degree

H. Which of the following best describes you current work status?

- ☐ Seeking Work
- ☐ Working full or part time (either outside the home or at a home-based business)
- ☐ Full time homemaker
- ☐ Not working due to my health
- ☐ Not working for other reasons

Section II. Quality of Life

Please indicate how your child has been feeling during the past two weeks by checking the box matching your response.

To what extent has your child had difficulty:	**A lot of difficulty**	**Some difficulty**	**A little difficulty**	**No difficulty**
1. Performing vigorous activities such as running or playing sports	☐	☐	☐	☐
2. Walking as fast as others	☐	☐	☐	☐
3. Climbing stairs as fast as others	☐	☐	☐	☐
4. Carrying or lifting heavy objects such as books, a school bag, or backpack	☐	☐	☐	☐
5. Climbing several flights of stairs	☐	☐	☐	☐

Please check the box matching your response.

*During the past **two weeks**, indicate how often your child:*	**Always**	**Often**	**Sometimes**	**Never**
6. Seemed happy	☐	☐	☐	☐
7. Seemed worried	☐	☐	☐	☐
8. Seemed tired	☐	☐	☐	☐
9. Seemed short-tempered	☐	☐	☐	☐
10. Seemed well	☐	☐	☐	☐
11. Seemed grouchy	☐	☐	☐	☐
12. Seemed energetic	☐	☐	☐	☐
13. Was absent or late for school or other activities because of his/her illness or treatments ……..	☐	☐	☐	☐

Please circle the number indicating your answer. Please choose only one answer for each question.

*Thinking about the state of your child's health over the past **two weeks**, indicate:*

14. The extent to which your child participated in sports and other physical activities, such as gym class
1. Has not participated in physical activities
2. Has participated less than usual in sports
3. Has participated as much as usual but with some difficulty
4. Has been able to participate in physical activities without any difficulty

15. The extent to which your child has difficulty walking
1. He or she can walk a long time without getting tired
2. He or she can walk a long time but gets tired
3. He or she cannot walk a long time, because he or she gets tired quickly
4. He or she avoids walking whenever possible, because it's too tiring for him or her

Please check the box that matches your response to these questions.

*Thinking about your child's state of health during the past **two weeks**, indicate the extent to which each sentence is true or false for your child:*

	Very true	Somewhat true	Somewhat false	Very false
16. My child has trouble recovering after physical effort	☐	☐	☐	☐
17. Mealtimes are a struggle	☐	☐	☐	☐
18. My child's treatments get in the way of his/her activities	☐	☐	☐	☐
19. My child feels small compared to other kids the same age	☐	☐	☐	☐
20. My child feels physically different from other kids the same age	☐	☐	☐	☐
21. My child thinks that he/she is too thin	☐	☐	☐	☐
22. My child feels healthy	☐	☐	☐	☐
23. My child tends to be withdrawn	☐	☐	☐	☐
24. My child leads a normal life	☐	☐	☐	☐
25. My child has less fun than usual	☐	☐	☐	☐
26. My child has trouble getting along with others	☐	☐	☐	☐
27. My child has trouble concentrating	☐	☐	☐	☐
28. My child is able to keep up with his/her school work or summer activities (e.g. camp)	☐	☐	☐	☐
29. My child is not doing as well as usual in school or summer activities (e.g. camp)	☐	☐	☐	☐
30. My child spends a lot of time on his/her treatments everyday…….	☐	☐	☐	☐

Please circle the number indicating your answer. Please choose only one answer for each question.

31. How difficult is it for your child to do his/her treatments (including medications) each day?
1. Not at all
2. A little
3. Moderately
4. Very

32. How do you think your child's health is now?
1. Excellent
2. Good
3. Fair
4. Poor

Section III. Symptom Difficulties

The next set of questions is designed to determine the frequency with which your child has certain respiratory difficulties, such as coughing or shortness of breath.

*Please indicate how your child has been feeling during the past **two weeks.***	**A great deal**	**Somewhat**	**A little**	**Not at all**
33. My child had trouble gaining weight	☐	☐	☐	☐
34. My child was congested	☐	☐	☐	☐
35. My child coughed during the day	☐	☐	☐	☐
36. My child had to cough up mucus	☐	☐	☐	☐ **Go to Question 38**

37. My child's mucus has been mostly: ☐ Clear ☐ Clear to yellow ☐ Yellowish-green ☐ Green with traces of blood ☐ Don't know

*During the past **two weeks:***	**Always**	**Often**	**Sometimes**	**Never**
38. My child wheezed	☐	☐	☐	☐
39. My child had trouble breathing	☐	☐	☐	☐
40. My child woke up during the night because he/she was coughing	☐	☐	☐	☐
41. My child had gas	☐	☐	☐	☐
42. My child had diarrhea	☐	☐	☐	☐
43. My child had abdominal pain	☐	☐	☐	☐
44. My child has had eating problems	☐	☐	☐	☐

Please be sure you have answered all the questions.

THANK YOU FOR YOUR COOPERATION!

CFQ-R

Adolescents and Adults **(Patients 14 Years Old and Older)**

CYSTIC FIBROSIS QUESTIONNAIRE - **REVISED**

Understanding the impact of your illness and treatments on your everyday life can help your healthcare team keep track of your health and adjust your treatments. For this reason, this questionnaire was specifically developed for people who have cystic fibrosis. Thank you for your willingness to complete this form.

Instructions: The following questions are about the current state of your health, as you perceive it. This information will allow us to better understand how you feel in your everyday life.

Please answer all the questions. There are **no** right or wrong answers! If you are not sure how to answer, choose the response that seems closest to your situation.

Section I. Demographics

Please fill-in the information or check the box indicating your answer.

A. What is your date of birth?

Date ____ ____ ____ ____ ____ ____ ____ ____
Mo Day Year

B. What is your gender?

☐ Male ☐ Female

C. During the **past two weeks**, have you been on vacation or out of school or work for reasons **NOT** related to your health?

☐ Yes ☐ No

D. What is your current marital status?

- ☐ Single/never married
- ☐ Married
- ☐ Widowed
- ☐ Divorced
- ☐ Separated
- ☐ Remarried
- ☐ With a partner

E. Which of the following best describes your racial background?

- ☐ Caucasian
- ☐ African American
- ☐ Hispanic
- ☐ Asian/Oriental or Pacific Islander
- ☐ Native American or Native Alaskan
- ☐ Other (please describe) _____
- ☐ Prefer not to answer this question

F. What is the highest grade of school you have completed?

- ☐ Some high school or less
- ☐ High school diploma/GED
- ☐ Vocational school
- ☐ Some college
- ☐ College degree
- ☐ Professional or graduate degree

G. Which of the following best describes your current work or school status?

- ☐ Attending school outside the home
- ☐ Taking educational courses at home
- ☐ Seeking work
- ☐ Working full or part time (either outside the home or at a home-based business)
- ☐ Full time homemaker
- ☐ Not attending school or working due to my health
- ☐ Not working for other reasons

Section II. Quality of Life

Please check the box indicating your answer.

*During the past **two weeks**, to what extent have you had difficulty:*	A lot of difficulty	Some difficulty	A little difficulty	No difficulty
1. Performing vigorous activities such as running or playing sports	☐	☐	☐	☐
2. Walking as fast as others	☐	☐	☐	☐
3. Carrying or lifting heavy things such as books, groceries, or school bags	☐	☐	☐	☐
4. Climbing one flight of stairs	☐	☐	☐	☐
5. Climbing stairs as fast as others	☐	☐	☐	☐

***During the past two weeks**, indicate how often:*	Always	Often	Sometimes	Never
6. You felt well	☐	☐	☐	☐
7. You felt worried	☐	☐	☐	☐
8. You felt useless	☐	☐	☐	☐
9. You felt tired	☐	☐	☐	☐
10. You felt energetic	☐	☐	☐	☐
11. You felt exhausted	☐	☐	☐	☐
12. You felt sad	☐	☐	☐	☐

Please circle the number indicating your answer. Please choose only one answer for each question.

*Thinking about the state of your health over the last **two weeks**:*

13. To what extent do you have difficulty walking?
1. You can walk a long time without getting tired
2. You can walk a long time but you get tired
3. You cannot walk a long time because you get tired quickly
4. You avoid walking whenever possible because it's too tiring for you

14. How do you feel about eating?
1. Just thinking about food makes you feel sick
2. You never enjoy eating
3. You are sometimes able to enjoy eating
4. You are always able to enjoy eating

15. To what extent do your treatments make your daily life more difficult?
1. Not at all
2. A little
3. Moderately
4. A lot

16. How much time do you currently spend each day on your treatments?
1. A lot
2. Some
3. A little
4. Not very much

17. How difficult is it for you to do your treatments (including medications) each day?
1. Not at all
2. A little
3. Moderately
4. Very

18. How do you think your health is now?
1. Excellent
2. Good
3. Fair
4. Poor

Please select a box indicating your answer.

*Thinking about your health during the past **two weeks**, indicate the extent to which each sentence is true or false for you.*	Very true	Somewhat true	Somewhat false	Very false
19. I have trouble recovering after physical effort	☐	☐	☐	☐
20. I have to limit vigorous activities such as running or playing sports	☐	☐	☐	☐
21. I have to force myself to eat	☐	☐	☐	☐
22. I have to stay at home more than I want to	☐	☐	☐	☐
23. I feel comfortable discussing my illness with others	☐	☐	☐	☐
24. I think I am too thin	☐	☐	☐	☐
25. I think I look different from others my age	☐	☐	☐	☐
26. I feel bad about my physical appearance	☐	☐	☐	☐
27. People are afraid that I may be contagious	☐	☐	☐	☐
28. I get together with my friends a lot	☐	☐	☐	☐
29. I think my coughing bothers others	☐	☐	☐	☐
30. I feel comfortable going out at night	☐	☐	☐	☐
31. I often feel lonely	☐	☐	☐	☐
32. I feel healthy	☐	☐	☐	☐
33. It is difficult to make plans for the future (for example, going to college, getting married, advancing in a job, etc.)	☐	☐	☐	☐
34. I lead a normal life	☐	☐	☐	☐

Section III. School, Work, or Daily Activities

Questions 35 through 38 are about school, work, or other daily tasks.

35. To what extent did you have trouble keeping up with your schoolwork, professional work, or other daily activities during the past **two weeks**?
1. You have had no trouble keeping up
2. You have managed to keep up but it's been difficult
3. You have been behind
4. You have not been able to do these activities at all

36. How often were you absent from school, work, or unable to complete daily activities during the last two weeks because of your illness or treatments?
☐ Always ☐ Often ☐ Sometimes ☐ Never

37. How often does CF get in the way of meeting your school, work, or personal goals
☐ Always ☐ Often ☐ Sometimes ☐ Never

38. How often does CF interfere with getting out of the house to run errands such as shopping or going to the bank?
☐ Always ☐ Often ☐ Sometimes ☐ Never

Section IV. Symptom Difficulties

Please select a box indicating your answer.

*Indicate how you have been feeling during the past **two weeks**.*	A great deal	Somewhat	A little	Not at all
39. Have you had trouble gaining weight?	☐	☐	☐	☐
40. Have you been congested?	☐	☐	☐	☐
41. Have you been coughing during the day?	☐	☐	☐	☐
42. Have you had to cough up mucus?	☐	☐	☐	☐ Go to Question 44

43. Has your mucus been mostly: ☐ Clear ☐ Clear to yellow ☐ Yellowish-green ☐ Green with traces of blood ☐ Don't know

*How often during the past **two weeks:***	Always	Often	Sometimes	Never
44. Have you been wheezing?	☐	☐	☐	☐
45. Have you had trouble breathing?	☐	☐	☐	☐
46. Have you woken up during the night because you were coughing?	☐	☐	☐	☐
47. Have you had problems with gas?	☐	☐	☐	☐
48. Have you had diarrhea?	☐	☐	☐	☐
49. Have you had abdominal pain?	☐	☐	☐	☐
50. Have you had eating problems?	☐	☐	☐	☐

Please be sure you have answered all the questions.

THANK YOU FOR YOUR COOPERATION!

APPENDIX I

Qualitative Survey Questionnaire

Qualitative Survey Questions

Directions: Thinking over the past thirty days, please answer the following questions as openly and honestly as you are able.

1. "If you have considered seeking mental health services, please identify specific barriers to seeking and receiving mental health treatment for yourself. Examples may include lack of time, finances, childcare, feelings of guilt/overwhelm/selfishness, CF and child are main focus/my issues are secondary and/or unimportant."
2. "If you are experiencing anxiety, please identify the areas in your life currently contributing to feelings of anxiety." Please note: anxiety is characterized as excessive worry, concern, fear in the present and future.
3. "If you are experiencing depression, please identify the areas in your life that contribute to feelings of depression." Please note: depression is characterized as feelings of sadness, feeling overwhelmed, "the blues," hopelessness, and helplessness.
4. “Please identify how your child's cystic fibrosis diagnosis has changed your life: past, present, and future. What are the greatest factors that contribute to your daily quality of life?" Please note: quality of life may be defined as when an individual is healthy, comfortable, and able to participate in or enjoy life events including physical, emotional, spiritual, material, and social wellbeing.
5. Many individuals within the CF community express their hope and excitement with the introductions of the CFTR modulator medications including: Kalydeco,

Orkambi, Symdeko, or Trikafta.

"If the introductions of these medications have changed your viewpoint, perspective, or relationship with CF, please describe how your thoughts and feelings are changing or evolving regarding CF in your current day to day life and going into the future?"

APPENDIX J

Informed Consent

INFORMED CONSENT

TEXAS WOMAN'S UNIVERSITY (TWU)
CONSENT TO PARTICIPATE IN RESEARCH

Title: An Exploration of the Experiences of Caregivers Raising Children with a Cystic Fibrosis Diagnosis and the Relationship Between Rates of Anxiety, Depression, and Quality of Life: An Online Mixed-Methods Study

Principal Investigator: Kristina Dingus, MA, MS, LMFT Associate
dingusresearch@gmail.com 972/841-9171

Faculty Advisor: Linda Ladd, PhD, PsyD lladd@twu.edu 940/898-2694

Summary and Key Information about the Study

You are being asked to participate in a research study for Mrs. Kristina Dingus at Texas Woman's University, as part of her dissertation. The purpose of the study is to explore the lived experiences of caregivers raising a child with a diagnosis of cystic fibrosis in order to better understand the quality of life, psychosocial functioning of the child (as reported by the caregiver), dealing with the child's health complications, and family and social struggles and other obstacles caregivers face on a daily basis, and the relationship between caregiver rates of anxiety, depression, and quality of life. The information gathered by this case study will be used to supplement the training mental health professionals receive on how to deliver higher quality therapeutic services to this demographic. You have been asked to participate in this study because you are a parent or caregiver of a child, between the ages of 1 day old and 18, diagnosed with Cystic Fibrosis. The greatest risks of this study include potential loss of confidentiality, loss of time, fatigue, emotional discomfort and/or emotional distress. We will discuss these risks and the rest of the study procedures in greater detail below.

Your participation in this study is completely voluntary. If you are interested in learning more about this study, please review this consent form carefully and take your time deciding whether or not you want to participate. Please feel free to ask the researcher any questions you have about the study at any time.

Description of Procedures

The total estimated time to complete this survey is 35 to 50 minutes. The length of time for each instrument is listed below and may also be found in Table 5.

As a participant in this study you will be asked to spend approximately three to five minutes reading about the proposed study and its purpose as well as completing this informed consent document.

As a participant in this study you will be asked to spend approximately five minutes of your time completing a demographic questionnaire for yourself and five minutes of your time completing a demographic/medical history questionnaire for your child in an online survey.

As a participant in this study you will be asked to spend approximately seven to fifteen minutes of your time completing assessments related to caregiver biopsychosocial (daily mental, emotional, physical, and social abilities) functioning, anxiety (GAD-7), depression (PHQ-9), quality of life (CQOLCF), and cystic fibrosis for yourself and your child in an online survey.

As a participant in this study you will be asked to spend approximately fifteen minutes of your time completing the Cystic Fibrosis Questionnaire-Revised (CFQ-R) on behalf of your child in an online survey. This questionnaire has recently began being distributed at routine clinic visits and you may already be familiar and previously completed. The CFQ-R measures functioning, anxiety, depression, quality of life, and CF-related complications.

As a participant in this study you will be asked to spend approximately fifteen minutes answering five open ended questions related directly to your individual and family experiences of raising a child with a cystic fibrosis diagnosis in an online survey.

The researcher will ask questions about your daily time commitment to assisting the individual with treatments, adherence to medical protocol, and the implications of your dedication to help the individual based on quality of life and biopsychosocial (daily mental, emotional, physical, and social abilities) functioning questionnaire measures. In order to be a participant in this study, you must be a caregiver of a child with a cystic fibrosis diagnosis between the ages of 1 day old to 18.

Please see the table below for a breakdown of the proposed measures and time commitment.

Description of Study Procedures

Study Component	Number of Questions	Length of Time to Complete
Reading about the Study & Informed Consent	N/A	3–5 minutes
Participant Demographic Questionnaire	27	3–5 minutes
Child Demographic Questionnaire	34	3–5 minutes
Personal Experience Question One	1	3 minutes
General Anxiety Disorder-7 (GAD-7)	7	1–2 minutes
Personal Experience Question Two	1	3 minutes

Patient Health Questionnaire-9 (PHQ-9)	9	1–3 minutes
Personal Experience Question Three	1	3 minutes
Caregiver Quality of Life Cystic Fibrosis (CQOLCF) Scale	35	5–10 minutes
Personal Experience Question Four	1	3 minutes
Cystic Fibrosis Questionnaire-Revised (CFQ-R)	Varies by age grouping ; no more than 45	5–15 minutes
Personal Experience Question Five	1	3 minutes

Potential Risks

If you are a parent or caregiver of a child with a cystic fibrosis diagnosis, the researcher will ask you questions about your daily time commitment to assisting your child with treatments, adherence to medical protocol, and the implications based on quality of life and psychosocial (daily mental, emotional, physical, and social abilities) functioning questionnaire measures. The researcher will also ask you questions about how having a child with a cystic fibrosis diagnosis has affected your relationships with your family and your friends. You will be asked to complete a series of questionnaires and assessments to quantify your individual levels of anxiety, depression, and quality of life.

1). A possible risk in this study is emotional discomfort and/or distress with the interview questions. If you become tired or upset you may take breaks as needed. You may also stop answering questions at any time and end the survey. The participant may withdraw from the study at any time. If you feel you need to talk to a professional about your discomfort, the researcher has provided a list of resources.

2). A possible risk in this study is fatigue. If you become tired or exhausted, you may take breaks as needed. You may also stop answering questions at any time and opt out of the online survey and study. If a question makes the participant uncomfortable, they will not be required to answer. The researcher will notify the participant that they may skip questions or withdraw from the study at any time.

3). A possible risk in this study is loss of confidentiality. Confidentiality will be protected to the extent that is allowed by law. At no time are you asked to identify your name, names of child, or such identifying information as your address. The study will ask for generalized information such as the type of CFTR mutation, and approximate ranges of FEV1/PFT, BMI, and weight, which cannot be traced back to you.

At the end of the main survey, participants will have the option of clicking on a link to go to a separate survey where they can provide their email address in order to receive an executive summary of the study results and/or to participate in a future focus group. The separate survey to collect email addresses will not be linked in any way to the main

survey. If you elect to provide your email, the data will be stored securely and will not be used, transferred, sold, or distributed to anyone outside of the scope of this study.

No one but the researcher will know your information. Only the researcher, a graduate student coder, and her major professor will have access to the online study. The results of the study will be reported in scientific magazines or journals and presented professionally; as noted above, neither your name or any other identifying information will be collected in the main study. There is a potential risk of loss of confidentiality in all email, downloading, electronic meetings and internet transactions.

4). A possible risk in this study is the loss of time spent completing the survey questions. Participants are notified they may take breaks at any time but the survey does not allow to save and return.

Participation and Benefits

Your involvement in this study is completely voluntary and you may withdraw from the study at any time. There are no direct benefits to you for volunteering to participate in the study. The overall benefit of this study is indirect to you, as it will contribute to the body of existing literature on Cystic Fibrosis, a rare, genetic, life-shortening illness that is understudied in the realm of psychological, mental, and behavioral health. The benefits of the study are to enhance the literature and knowledge in the field. If you would like to know the results of this study, we will email them to you at the email address you voluntarily provide by clicking on the link to a new website, located at the end of this survey.

The researchers will try to prevent any problem that could happen because of this research. You should let the researchers know at once if there is a problem and they will help you. However, TWU does not provide medical services or financial assistance for injuries that might occur because you are taking part in this research.

Questions Regarding the Study

If you have any questions about the research study you should ask the researchers; their contact information is at the top of this form. If you have questions about your rights as a participant in this research or the way this study has been conducted, you may contact the TWU Office of Research and Sponsored Programs at 940-898-3378 or via e-mail at IRB@twu.edu.

By checking this box, you are electronically providing your signature and today's date.

Psychological, Behavioral, and Mental Health Referral List:
AAMFT Therapist Locator: http://aamft.org
APA Therapist Locator: http://www.apa.org

Open Path Psychotherapy Collective: http://www.openpathcollective.org
Psychology Today: http://www.therapists.psychologytoday.com
Therapist Locator: http://www.therapistlocator.net

APPENDIX K

Participant Survey Instructions

The following information will be provided to the participants after they have read the informed consent and provided their electronic signature.

Participant Survey Instructions

Survey and Instructions

Instructions are provided throughout the survey to guide you in each section. You may withdraw from the survey at any time without penalty.

The following information is an outline of the survey format so you are aware of what is coming and what to expect in each section:

1. The Participant Caregiver Demographic Questionnaire. This will take approximately five minutes to complete. You will be asked to answer questions related to your age, sex, gender, education, employment information, and mental health.

2. The Child Demographic Questionnaire. This will take approximately five minutes to complete. You will be asked to answer questions related to your diagnosed child's education, physical health characteristics including CFTR gene mutation, FEV_1/PFT, BMI, hospitalizations, mediations, mental health, etc...

3. Personal Experience Question One is an open-ended format and will take approximately three minutes to answer. Please keep in mind the length of the overall survey to avoid exhaustion, fatigue, and survey burnout.

4. The General Anxiety Disorder-7 (GAD-7) Questionnaire. There are seven questions and will take you one to two minutes to complete. This assessment will ask you to identify how frequently you have exhibited feelings of anxiety (irritability, worry, nervousness, etc...) in the past two weeks.

5. Personal Experience Question Two is an open-ended format and will take approximately three minutes to answer.

6. The Patient Health Questionnaire-9 (PHQ-9). There are nine questions and will take less than three minutes to complete. This assessment will ask you to identify how frequently you have exhibited feelings of depression (difficulty sleeping, change in appetite, and trouble concentrating) in the past two weeks.

7. Personal Experience Question Three is an open-ended format and will take approximately three minutes to answer.

8. The Caregiver Quality of Life Cystic Fibrosis (CQOLCF) Scale. There are 35 questions and will take you approximately 5 to 10 minutes to complete. This assessment will ask you to rate your satisfaction in various areas of your life (routine, support, and communication) over the past week.

9. Personal Experience Question Four is an open-ended format and will take approximately three minutes to answer.

10. The Cystic Fibrosis Questionnaire-Revised (CFQ-R). This will take you approximately 15 minutes to complete. This assessment will ask you to answer questions related to your diagnosed child's daily physical, emotional, and mental functioning (ability to climb stair, ability to sleep, disruption of treatments on fun activities) over the past two weeks.

11. Personal Experience Question Five is an open-ended format and will take approximately three minutes to answer.

A link to separate survey to voluntarily enter your email address if you would like an executive summary of the study results and/or if you would like to be contacted in the future regarding mental health focus groups or study participation.

This is the end of the survey. Thank you for your participation! If you feel you need additional support, resources are provided below.

Psychological, Behavioral, and Mental Health Referral List:

AAMFT Therapist Locator: http://aamft.org

APA Therapist Locator: http://www.apa.org

Open Path Psychotherapy Collective: http://www.openpathcollective.org

Psychology Today: http://www.therapists.psychologytoday.com Therapist Locator: http://www.therapistlocator.net

APPENDIX L

Descriptive Statistics Table

Descriptive Statistics Tables

Caregiver Variables: Definition, Type, Level of Measurement

Variable	Type of Variable	Level of Measurement	Definition	Assessment
Gender	Independent	Nominal	Biological Sex M/F	Self-report
Age	Independent	Ratio	Chronological Age	Self-report
Race	Independent	Nominal	Racial Background	Self-report
Ethnicity	Independent	Nominal	Ethnic Identity	Self-report
Marital Status	Independent	Nominal	M/S/D/W/Cohab	Self-report
Level of Education	Independent	Ordinal	Level of academic attainment: No HS, GED, HS, some college B, M, Phd/MD	Self-report
Employment Status	Independent	Nominal	PRN, PT, FT, SAHC, UN	Self-report
Insurance Coverage	Independent	Nominal	Yes/No	Self-report
Number of Children	Independent	Ratio	Number of total children	Self-report
Number of Children with CF	Independent	Ratio	Number of children with CF	Self-report
Age of Children	Independent	Ratio	Chronological Age of child	Self-report
Age of Children with CF	Independent	Ratio	Chronological age of child	Self-report
Religious Affiliation	Independent	Nominal	Religious/Spiritual Beliefs	Self-report
Anxiety	Independent	Nominal	Diagnosed with anxiety Y/N	Self-report
Depression	Independent	Nominal	Diagnosed with depression Y/N	Self-report
Other mental health illness diagnosis	Independent	Nominal	Diagnosed with other MH Y/N	Self-report

Mental health treatment	Independent	Nominal	Yes, No, Current, Previous	Self-report
Psychotropic Medication	Independent	Nominal	Yes, No, Current, Previous	Self-report
Caregiver Quality of Life	Dependent	Interval	The quality of life as reported by the caregiver	Caregiver Quality of Life-Cystic Fibrosis (CQOLCF)
Anxiety	Dependent	Interval	Level of anxiety reported by caregiver	General Anxiety Disorder-7 (GAD-7)
Depression	Dependent	Interval	Level of depression reported by caregiver	Patient Health Questionnaire-9 (PHQ-9)

- Limitations: all self-report; physical health data may not be 100% accurate

Child Variables: Definition, Type, Level of Measurement

Variable	Type of Variable	Level of Measurement	Definition	Assessment
Age	Independent	Ratio	Chronological age	Self-report
Age of CF Diagnosis	Independent	Ratio	Age when child was diagnosed with CF	Self-report
Gender	Independent	Nominal	Biological sex male/female	Self-report
CF Gene Mutation 1	Independent	Nominal	Category of CF	Self-report
CF Gene Mutation 2	Independent	Nominal	Category of CF	Self-report
FEV_1/ (PFT)	Independent	Ratio	Forced expiratory volume in 1 second, a measure of pulmonary function (Boling et al., 2003)	Self-report

			Pulmonary function test	
BMI	Independent	Ratio	Body Mass Index	Self-report
Weight	Independent	Ratio	Weight of child	Self-report
CF Severity	Independent	Ordinal	Mild, Moderate, Severe (as dx by PCP)	Self-report
Lung Transplant	Independent	Nominal	Yes/No	Self-report
Lung Transplant Waiting List	Independent	Nominal	Yes/No	Self-report
Co-morbid diagnosis (diabetes, asthma, other)	Independent	Nominal	Additional diagnosis of child with CF	Self-report
Tune-up within the past 6 months	Independent	Nominal	Routine tune-up needed to slow disease progression and maintenance	Self-report
Nasal surgery within the past 6 months	Independent	Nominal	Surgery needed to clear mucous from airways	Self-report
Positive *Pseudomonas aeruginosa* culture	Independent	Nominal	Presence of pseudamonas bacteria	Self-report
Positive M*ycobacterium abscessus* culture	Independent	Nominal	Presence of Mycobacterium bacteria	Self-report
Positive *Burkholderia cenocepacia* culture	Independent	Nominal	Presence of B. Cepacia bacteria	Self-report
Anxiety	Independent	Nominal	Child diagnosed with anxiety Y/N	Self-report

Depression	Independent	Nominal	Child diagnosed with depression Y/N	Self-report
Other mental health illness diagnosis	Independent	Nominal	Child diagnosed with other MH Y/N	Self-report
Mental health treatment	Independent	Nominal	Yes, No, Current, Previous	Self-report
Child Education Status (Traditional, Montessori, Homeschool, Hybrid)	Independent	Nominal	Type of school the child attends	Self-report
Psychotropic Medication	Independent	Nominal	Yes, No, Current, Previous	Self-report
Child Quality of Life	Dependent	Interval	The child's quality of life as perceived and reported by the caregiver.	Cystic Fibrosis Questionnaire-Revised (CFQ-R) Self-report

- Limitations: all self-report; physical health data may not be 100% accurate

APPENDIX M

Request for Caregiver Quality of Life Cystic Fibrosis Scale

TWU

Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu>

CQOLCF

Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu> Thu, Apr 2, 2020 at 1:28 PM
To: Whitney Boling <Whitney.Boling@indstate.edu>

Hello Dr. Boling!

I hope this email finds you well. In 2018 you granted me permission to use the CQOLCF in a case study I was conducting. I wanted to reach out to you again to obtain permission to use the CQOLCF in my dissertation research.

The dissertation is titled, "An Exploration of the Experiences of Caregivers Raising Children with a Cystic Fibrosis Diagnosis and the Relationship between rates of Anxiety, Depression, and Quality of Life: An Online Exploratory Mixed-Methods Study."

This is a mixed-methods approach with a series of four qualitative questions to better understand the psychological needs of this population. The quantitative measures being used are the Patient Health Questionnaire-9 (PHQ-9), General Anxiety Disorder-7 (GAD-7), Cystic Fibrosis Questionnaire- Revised (CFQ-R), and CQOLCF.

I am happy to provide any additional information you may need in consideration of granting permission to use the CQOLCF Scale. Please let me know if you have any questions or concerns!

I look forward to hearing from you.

In Gratitude,

Kristina Dingus, MA, MS, LMFT Associate

[Quoted text hidden]

TWU

Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu>

CQOLCF

Whitney Nesser <Whitney.Nesser@indstate.edu> Fri, Apr 3, 2020 at 9:27 AM
To: Kristina Dingus Keuhlen <kdinguskeuhlen@twu.edu>

Hello Kris*na,

Thank you so much for contac*ng me again! I hope this email finds you well too.

Congratula*ons on your progression to disserta*on! Yes, you may certainly use the CQOLCF, and I am happy to help in any way you want/need.

Also, I have remarried (hence, last name Nesser instead of Boling), but please s*ll use the same CQOLCF cita*on - the one with Boling.

Please let me know if you have any ques*ons, and wishing you the best of luck!

Sincerely,

Whitney

Whitney Nesser, PhD, MBA, CHES

Associate Dean for Academics

College of Health and Human Services

Indiana State University

401 N. 4th Street, HHS 403

(812) 237-3105/3114 | whitney.nesser@indstate.edu

STATE

APPENDIX N

Request for Cystic Fibrosis Questionnaire-Revised

Gmail Kristina Dingus <kristinadingus@gmail.com>

CFQ-R Permissions Request for Dissertation

Kristina Dingus <kristinadingus@gmail.com> Thu, Feb 13, 2020 at 1:14 PM
To: Alexandra.Quittner@nicklaushealth.org

Hello Dr. Quittner,

My name is Kristina Dingus Keuhlen and I am a Doctoral Candidate at Texas Woman's University. I am in the process of writing my dissertation proposal. I am a Marriage and Family Therapist by day and my dissertation is seeking to understand the experiential and biopsychosocial factors contributing to caregiver barriers to seeking and receiving mental health care and the relationship between rates of anxiety, depression, and quality of life.

This is a mixed-methods approach with a series of four qualitative questions to better understand the psychological needs of this population. The quantitative measures being used are the Patient Health Questionnaire-9 (PHQ-9), General Anxiety Disorder-7 (GAD-7), and Caregiver Quality of Life Cystic Fibrosis (CQOLCF) Scale. I also wanted to incorporate the CFQ-R into the study to identify the overall health, wellness, and symptoms of the diagnosed child, as reported by the caregiver, to explore the connection there as well.

I would like to request permission to use the CFQ-R and access to scoring and administration manuals in my online, mixed-methods dissertation study. Please let me know if you need any additional information from me or if I need to go a different route to request access.

I look forward to hearing from you.

--
In Gratitude,

Kristina Dingus Keuhlen, MA, MS, LMFT Associate
Texas Woman's University Marriage and Family Therapy Doctoral Candidate
www.northtexasfamilytherapy.com

Gmail Kristina Dingus <kristinadingus@gmail.com>

Fwd: CFQ-R English US and UK

Alexandra Quittner <aquittner0202@gmail.com> Mon, Apr 6, 2020 at 7:50 AM
To: Kristina Dingus <kristinadingus@gmail.com>
Cc: Alexandra Quittner <Aquittner0202@gmail.com>, Alexandra Quittner <alexandra.quittner@nicklaushealth.org>

Hi Kristina
Here are the instruments…just sending along all of the versions

Begin forwarded message:

From: Alexandra Quittner <aquittner0202@gmail.com>
Subject: CFQ-R English US and UK
Date: April 6, 2020 at 8636657 AM EDT
To: krisindingus@gmail.com
Cc: Alexandra Quittner <Aquittner0202@gmail.com>, Alexandra Quittner <alexandra.quittner@nicklaushealth.org>

Hi Kristina
Here is the CFQ-R Teen Adult version in US English and UK in case you need it.

Alexandra

Dr. Alexandra Quittner
305 992-2411

2 attachments

All Versions of the UK CFQ-R Final.pdf
165K

All Versions English CFQ-R FINAL.pdf
151K

APPENDIX O

Copyright Agreement for the CFQ-R

Copyright Agreement for the CFQ-R

CYSTIC FIBROSIS QUESTIONNAIRES - REVISED

Copyright Agreement for the Cystic Fibrosis Questionnaires – Revised

Alexandra L. Quittner, Ph.D.

The undersigned agrees:

1. to administer and score the CFQ-R for research or clinical purposes only,
2. not to alter the items or scoring of the CFQ-R in any way,
3. to acknowledge the authors of the CFQ R in any publications that include the use of the CFQ-R,
4. not to distribute the CFQ-R to anyone without the explicit, written permission of the copyright holders,
5. not to administer or score, or in any way provide services related to the CFQ-R to a third party, for a fee without the explicit, written permission of the copyright holders,
6. that permission for the individual/organization named below to use the CFQ-R may be rescinded by the copyright holders at any time.

03/13/2020

Kristina Dingus Keuhlen, MA, MS, LMFT Associate kdinguskeuhlen@twu.edu
Texas Woman's University, 304 Administration Drive, Denton, Texas 76204

Please sign, scan and email to: Dr. Alexandra L. Quittner: aquittner0202@gmail.com
(305) 992-2411