THE IMPACT OF PARENTAL PSYCHOLOGICAL DISTRESS AND FUNCTIONING ON CHILDREN WITH CYSTIC FIBROSIS: ASSESSING ADHERENCE, LUNG FUNCTION, AND QUALITY OF LIFE

By

ALYSSA M. FRITZ

A DISSERTATION PRESENTED TO THE GRADUATE SCHOOL OF THE UNIVERSITY OF FLORIDA IN PARTIAL FULFILLMENT OF THE REQUIREMENTS FOR THE DEGREE OF DOCTOR OF PHILOSOPHY

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THE IMPACT OF PARENTAL PSYCHOLOGICAL DISTRESS AND FUNCTIONING ON CHILDREN WITH CYSTIC FIBROSIS: ASSESSING ADHERENCE, LUNG FUNCTION, AND QUALITY OF LIFE

By

Alyssa M. Fritz

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Major: Psychology

Cystic fibrosis is one of the most challenging pediatric illnesses for children and their families to manage. The stressful and time-consuming treatments associated with cystic fibrosis may give rise to psychological sequelae in both children and their parents. Parents of children with CF are at-risk for mental health distress and ineffective coping behavior. These stressors have the potential to influence their children’s medical treatment, as well as contribute to downstream mental health challenges. Due to the paucity of information, this study assessed the impact of parental psychological distress on children’s morbidity, treatment adherence, and health-related quality of life (HRQOL). Parents completed depression and anxiety questionnaires, and reported ways they cope with common challenging cystic fibrosis-specific situations. Child adherence to CF treatments was measured using an empirically validated 24-hour recall and morbidity indices were collected from medical record review. Twenty-five parent-child dyads between 4-12-years-old completed the study across two sites (University of Florida and Virginia Commonwealth University).
Findings indicate a significant proportion of parents report mild to severe symptoms of depression and/or anxiety. Results revealed that effectiveness of parent-reported coping behavior is associated with lung function \( F(1, 22) = 8.07, p = .01, R^2 = .268, \hat{f}^2 = .367 \) and HRQOL for physical symptoms \( F(1, 22) = 10.533, p = .004, R^2 = .324, \hat{f}^2 = .479 \). Parent-reported symptoms of depression and anxiety were not associated with child health outcomes (adherence, lung function, HRQOL). Additional results were discussed in terms of effect sizes and clinical differences.
CHAPTER 1
INTRODUCTION

Pediatric Cystic Fibrosis

Cystic fibrosis (CF) is a chronic condition affecting approximately 30,000 people nationwide and 100,000 globally. Nearly 1,000 new cases are diagnosed annually in the United States (Cunningham & Taussig, 2013). This genetically inherited disease affects the lungs, pancreas, and reproductive organs. Diagnosis for CF most commonly occurs shortly after birth via newborn screening and a subsequent sweat test (Cunningham & Taussig, 2013). Current median survival age for individuals with CF is 40.7-years-old (Cystic Fibrosis Foundation Annual Report, 2014).

There are more than 1,800 different gene mutations in CF, with various mutations associated with disease symptoms and severity profiles (Cunningham & Taussig, 2013; Cystic Fibrosis Foundation, 2014). Individuals must inherit two copies of the defective gene, which subsequently produces an abnormal cystic fibrosis transmembrane conductance regulator (CFTR) protein that causes mucus buildup affecting the pancreas, the liver bile duct, the male reproductive system, and the lungs (Cystic Fibrosis Foundation Annual Report, 2014). Individuals diagnosed with CF manage gastrointestinal (GI) challenges, including poor nutrient absorption subsequent to the mucus preventing the release of pancreatic digestive enzymes. This combination of factors can result in growth deficiencies and potential malnutrition. Further, liver disease may result if mucus blocks the bile duct in the liver. The respiratory problems are typically the most persistent challenges faced by these individuals (Cystic Fibrosis Foundation Annual Report, 2014). When mucus obstructs airways, the resulting
inflammation and infections can lead to chronic lung disease and necessitate lung transplant in some individuals.

**Cystic Fibrosis Treatment and Management:** CF treatment regimens are particularly demanding. Completion of all recommended treatments is intensive and estimated to take two to four hours per day (Sawicki et al., 2011). There are multiple steps involved for completing the recommended treatment protocol. Current standard medical recommendations for individuals with CF include respiratory care, infection prevention and control, nutrition and GI care, and fitness recommendations. Respiratory care requires inhaled medications (e.g., bronchodilators) to open airways; mucolytics to thin mucus and improve clearance from the lungs; choosing an airway clearance technique (e.g., chest physical therapy, high-frequency chest wall oscillation, huff coughing) to loosen and dislodge mucus from airways and then clearing mucus through coughing or huffing (Cunningham & Taussig, 2013; Cystic Fibrosis Foundation, 2014). These treatments last between 20-40 minutes and occur multiple times per day, typically early morning and prior to bed (Cystic Fibrosis Foundation, 2014). Ensuring completion of daily respiratory treatments reduces lung infections and improves lung function.

Additional recommendations focus on nutrition and fitness concerns. Nutrition recommendations address the potential difficulty for absorbing nutrients due to mucus build up. Youth with CF are asked to consume 120% to 150% of the recommended dietary allowance and take enzymes with meals and snacks in order to promote growth and functioning (Cunningham & Taussig, 2013; Cystic Fibrosis Foundation, 2014). CF fitness recommendations are important for lung function, as well as strengthening
bones, loosening mucus, encouraging coughing, managing CF related diabetes and heart disease, and improving mood (Cunningham & Taussig, 2013). The combination of these treatment recommendations is time consuming, requires scheduling and places additional burdens on the family.

Life expectancy for individuals with CF has improved; however, adherence to these numerous time-consuming treatment regimens remains challenging. Non-adherence to CF treatment regimens is widespread and associated with increased symptom frequency, more hospitalizations, greater stress and family conflict, increased health care utilization and cost, as well as increased likelihood for morbidity and mortality (Barker & Quitter, 2010; Modi & Quittner, 2006; Modi, Lim, Geller, Wagner, & Quittner, 2006). Patient adherence to treatment recommendations varies across therapies and is typically poor (Modi, Lim, Yu, Gellar, Wagner, & Quittner, 2006; DiGirolamo, Quittner, Ackerman, & Stevens, 1997). Findings for adherence to respiratory treatments ranges from 40-90% (DeLambo, Levers-Landis, Drotar, & Quittner, 2004; Eakin & Riekert, 2013; Levers et al., 1999; Modi et al., 2006; Zindani, Streetman, & Nasr, 2006). Adherence to nebulized medication was 48% in one study, while airway clearance treatment is often problematic with ranges between 40-51% in other studies (Llorente, Garcia, & Martin, 2008; Modi, 2006; Quittner, Drotar, & Levers-Landis, 2000). DeLambo and colleagues (2004) reported that airway clearance and nutrition recommendations were associated with the most barriers, while other studies found that up to 92% of families report barriers to completing airway clearance treatments (Modi & Quittner, 2006). When completing daily structured adherence assessments, more than 50% of youth with CF reported completing less than the
prescribed airway clearance treatments, while approximately 30% stated that they were not completing any prescribed treatment (DiGirolamo, Quittner, Ackerman, & Stevens, 1997). Beyond the extensive logistical and time barriers, parents frequently report child oppositional behavior as interfering with treatment completion (Modi & Quittner, 2006). Parents are required to manage the logistical organization associated with scheduling treatments, the significant time requirements for treatment completion, as well as frequent child opposition in order to attempt to adhere to treatment recommendations.

**The Role of Parents in Cystic Fibrosis**

Parents are a central component of the family system during childhood and can impact child health outcomes. They complete global parenting tasks, including provision of emotional support, managing schedules, and implementing discipline. However, parents of youth with a chronic illness must complete additional illness-specific tasks, including daily components of treatment management and challenges associated with implementation of the treatment regimen timeline and scheduling (Ievers & Drotar, 1996; Quittner, DiGirolamo, Michel, & Eigen, 1992). The integral role of parents in the context of a pediatric chronic illness is illustrated in the social ecological model of child adjustment. This model posits that parent and child functioning is a transactional relationship influenced by a number of variables across the family system and beyond (e.g., coping, illness characteristics, the broad sociocultural context; Kazak et al., 2009). According to this model, parent functioning is closely interrelated to child adjustment and can influence child treatment outcomes.

Extant research indicates a robust relationship between parent functioning and a variety of child outcomes including pulmonary functioning, distress and internalizing.
symptoms, and quality of life (Fedele et al., 2013; Patterson, McCubbin, & Warwick, 1990; Quittner et al., 1996; Quittner, Drotar, & Ievers, 1998; Ryan et al., 2010). Crosssectional research has found relationships between elevated parent and child distress ratings, poorer family functioning related to poorer adherence to medication, and maternal depression symptoms related to poorer medication adherence (Besier & Goldbleck, 2011; DeLambo et al., 2004; Everhart et al., 2014; Ryan et al., 2010; Smith, Modi, Quittner, & Wood, 2010; Quittner, Barker, & Gellar, 2007; Quittner et al., 2014). A recent longitudinal study indicates that children of parents with elevated parent depression or anxiety symptoms are twice as likely to self-report symptoms of depression or anxiety (Quittner et al., 2014). Additional longitudinal studies found that measures of maternal stress predicted child adjustment symptoms (Fedele et al., 2013) while temporal precedence of parent distress has been related to subsequent child distress (Ryan et al., 2010). Other findings are indicate longitudinal indirect relationships between self-reported parent stress and coping and variance in child height, weight, and pulmonary functioning (Patterson, McCubbin, & Warwick, 1990).

Parents have an integral role in treatment thereby making the examination of the role of parental psychosocial functioning and management strategies on child health-related outcomes a necessary endeavor. Within CF, parents are responsible for integrating illness-specific care into the family routine. Care for children with CF involves time-consuming and multifaceted medical treatment regimens, including medications, diet management, and breathing treatments (Barker & Quittner, 2010). Specifically, parents manage complex treatment regimens, schedule daily airway clearance treatments, ensure children successfully complete multistep treatment processes (e.g.,
bronchodilator, mucolytics, airway clearance, and antibiotic), prescription refills, doctors’ appointments, make appropriate nutrition choices, as well as endure additional financial and personal stress associated with having a child with chronic illness (Besier, Born, Henrich, Hinz, Quittner, Goldbeck & the TIDES group, 2011; Wysocki & Gavin, 2006; Butcher & Nasr, 2015). Due to this high level of parental involvement, it is anticipated that parent functioning will affect children’s functioning levels. These demands may exacerbate stressors and place parents at risk for implementing less effective coping behaviors and increased psychological distress. Impaired family functioning may place children at risk for decreased medication adherence, poorer lung function, and poorer health related quality of life (Everhart, Fiese, Smyth, Borschuk, & Anbar, 2014; Quittner, Opipari, Regoli, Jocobsen, & Eigen, 1992; Quittner, Espelage, Opipari, Carter, Eid, & Eigen, 1998).

**Parental Psychosocial Factors**

In response to increased awareness of the demanding treatment regimen as well as the transactional relationship between parent mental health and childhood health outcomes, parental mental health and coping behaviors are emerging areas of research in cystic fibrosis. The following sections will outline the importance of parental mental health factors and psychological distress, specifically focusing on depression and anxiety. Specifically, this will assess the impact of parent symptoms on child outcomes of disease morbidity (e.g., lung function), treatment adherence, and child HRQOL.

**Depression and Anxiety**

Psychological distress is common in parents of CF-diagnosed youth. Recently, an international collaboration of researchers completed The International Depression Epidemiological Study (TIDES) across Europe (Quittner et al., 2014). TIDES was
conducted with parents of children at birth through 18-years-old diagnosed with CF as well as adolescents with CF 12-18-years-old. This study found elevated levels of depression in 37% of mothers and 31% of fathers as well as anxiety elevations in 48% of mothers and 36% of fathers using the measures proposed in current study (Quittner et al., 2014). Another study compared parents of CF patients to a control group sample, with results indicating that 28% of CF patient parents reported clinically significant symptoms of depression versus 21% in the control group and 17% of endorsed clinically significant symptoms of anxiety versus approximately 5% in the general population (Besier et al., 2011). Across studies, symptoms of depression in parents of youth with CF are estimated to range from 23% for fathers to 35% for mothers whereas anxiety symptoms range from 32% for fathers to 51% for mothers (Besier et al., 2011; Driscoll, Montag-Leifling, Acton, & Modi, 2009; Quittner et al., 2014). If parents reported symptoms of anxiety, they were 13.64 times more likely to report elevated symptoms of depression, with mothers being 15.52 times more likely to report depression symptoms (Quittner et al., 2014).

Increased symptoms of depression and anxiety among parents are associated with parental and youth maladjustment. Parents with higher symptoms of depression and anxiety experience increased risk factors, including increased levels of stress, and report reduced health-related quality of life (HRQOL) or life satisfaction (Besier et al., 2010; Driscoll, Montag-Leifling, Acton, & Modi, 2009; Driscoll et al., 2010). The impact of parental psychological distress on child functioning and health-related outcomes in CF has been examined in a small number of studies. Maternal depression was associated with rates of 11% less adherence to enzyme treatments and symptoms of
parent depression have also been associated with poorer lung function (Modi, Driscoll, Montag-Leifling, & Acton, 2011). Other studies have reported that parent depression was associated with poorer adherence to enzyme adherence and lack of weight gain between clinic visits (Quittner, Barker, Gellar, Butt, & Gondor, 2007). Notably, a transactional relationship exists between parent and youth psychological distress such that children of parents who report co-occurring anxiety or depression are more likely to report increased symptoms of both anxiety and depression themselves (Quittner et al., 2014). This link is especially important given that symptoms of anxiety and depression in youth with CF are associated with poorer treatment adherence and poorer clinic attendance (Smith, Modi, Quittner, & Wood, 2010; Snell et al., 2014; Quittner et al., 2014).

**Parental Coping**

As previously noted, parents of children with CF are at-risk for poorer overall psychosocial functioning. Assessing the effectiveness of their coping behaviors provides important information regarding specific disease management strategies and general behavioral strategies used to handle stressors. Coping as a construct refers to the combination of cognitive and behavioral efforts utilized to manage stressful situations (Lazarus & Folkman, 1984). Within CF, parents must continuously manage numerous disease-related problems, including the management of both the daily components of treatment as well as the challenges associated with implementation of treatment regimen timeline. They play an integral role in shaping children’s health behavior habits from a very young age and are responsible for medication and CF regimen adherence, diet, and activity levels. Given the complex treatment demands of CF, parent coping effectiveness affects child disease management (Patterson, McCubbin, & Warwick,
Using self-report measures, studies found associations between parent coping behavior and child health outcomes, including pulmonary function and height and weight changes. Less effective strategies were associated with poorer parent and child adjustment. Coping behaviors can range from implementing effective and adaptive strategies to utilizing ineffective or maladaptive strategies. Caring for a child with CF requires implementing a variety of coping behaviors across multiple domains, including behaviors to ensure treatment adherence, discipline strategies, response to medical procedures or symptoms, as well as spousal issues. Research shows that these parents are already at increased risk for depression and anxiety symptoms, which in turn can negatively impact family functioning and disease management (Besier et al., 2011; Quittner et al., 2014).

Parental ability to manage multiple, complex, and time-consuming treatments are integral for the health outcomes of their children (Thompson et al. 1992; Eddy et al. 1998; Graetz et al. 2000; Anderson et al. 2001; Berge & Patterson 2004; DeLambo et al. 2004). School-age children rely on their parents for managing medication adherence, airway clearance treatments, medical appointments, prescription refills, diet and nutrition options, with younger children requiring even greater assistance. Previous research in CF relies on parental self-reporting of coping via questionnaires (Patterson,McCubbin, & Warwick, 1990; Wong & Heriot, 2008). This method does not allow for assessment of the effectiveness of a coping strategy within the implementation context (Quittner & DiGirolamo, 1998; Stone, Greenberg, Kennedy-Moore, & Newman, 1992). There has been minimal research assessing links between stressful recalled situations and endorsed coping behavior (Quittner & DiGirolamo, 1998). Using empirical criteria to
assess the efficacy of a coping response allows for individualized assessment of family responses, particularly regarding domains of strengths and weaknesses (Quittner & DiGirolamo, 1998). Evaluating strategies based on their capacity to decrease the frequency, or perceived difficulty, of relevant problems provides the ability to link parent-generated strategies to the precipitating problem to provide specific, empirically-derived criteria for judging coping effectiveness, and links coping behavior directly to problems. Parents’ use of coping behavior impacts the effectiveness of illness-specific treatment integration in family functioning. Therefore, by assessing parent mental health and integration of coping behavior in response to both disease-specific and typical family functioning, this study provided specific information regarding daily disease management decision-making processes.

**Limits of the Current Literature**

A burgeoning body of research indicates that parents of youth with CF are at-risk for increased psychological distress and implementing less effective coping behavior. A limited number of studies have examined the relationship between parental psychological distress, coping, and child health outcomes. Clear gaps remain in the current literature on the impact of the relationship between parental symptoms of depression and anxiety, coping behavior, child treatment outcomes and child HRQOL. This current study aims to fill a number of important gaps in the CF literature, including utilization of objective medical outcomes (i.e., lung function, and detailed daily adherence information via a validated 24-hour recall) to determine the relations between parent-reported symptoms of depression and anxiety on child treatment outcomes. Additionally, this study builds on previous coping literature by utilizing an externally valid
measure in order to determine CF-specific coping behavior associated with common disease-management and family-functioning situations. Therefore, the purpose of this study was to assess the relations among parent reported symptoms of depression, anxiety, and coping behaviors on child health outcomes (e.g., adherence, lung function, HRQOL in 4-12 year-olds diagnosed with cystic fibrosis. This age group was chosen due to importance of parent management for treatment-related behaviors in this age group.

**Aims and Hypotheses**

**Primary**

**Aim 1:** Examine whether parent mental health at baseline was associated with child medication adherence, lung function, and HRQOL at baseline.

- **Hypothesis 1a:** Parents reporting higher symptoms of depression and anxiety were hypothesized to have children with lower treatment adherence.
- **Hypothesis 1b:** Parents reporting higher symptoms of depression and anxiety were hypothesized to have children with poorer lung function at baseline.
- **Hypothesis 1c:** Parents reporting higher symptoms of depression and anxiety were hypothesized to have children with lower HRQOL at baseline.

**Aim 2:** Examine whether effectiveness of parent coping behaviors was associated with child disease morbidity, treatment adherence, and HRQOL at baseline.

- **Hypothesis 2a:** Parents reporting less effective coping behaviors were hypothesized to have children with lower treatment adherence at baseline.
- **Hypothesis 2b:** Parents reporting less effective coping behaviors were hypothesized to have children with poorer lung function at baseline.
- **Hypothesis 2c:** Parents reporting less effective coping behaviors were hypothesized to have children with lower rated HRQOL at baseline.
Secondary

Aim 1: Compare child outcomes (i.e., adherence, lung function, HRQOL) between parents who reported depressive symptoms with mild and moderate clinical elevations to parents who reported subclinical symptoms at baseline.

Hypothesis 1a: Parents reporting both mild and/or moderate clinical elevations of depression were hypothesized to have children with poorer outcomes compared to children of parents who report subclinical symptoms.

Hypothesis 1b: Parents reporting both mild and/or moderate clinical elevations of anxiety were hypothesized to have children with poorer outcomes compared to children of parents who report subclinical symptoms.

Aim 2: Examine if the relationship between parent mental health and child health outcomes was mediated by effectiveness of parent coping behaviors at baseline.

Hypothesis 2a: Effectiveness of parent coping behavior was hypothesized to mediate the relationship between parent mental health symptoms and child health outcomes.

Exploratory Aim 1: Examine whether parent mental health was associated with child disease morbidity, medication adherence, and HRQOL at six-month follow-up.

Hypothesis 1a: Parents reporting more symptoms of depression and anxiety were hypothesized to have children with poorer outcomes at follow-up.

Exploratory Aim 2: Determine whether effectiveness of parent coping behaviors was associated with child disease morbidity, medication adherence, and HRQOL at six-month follow-up.

Hypothesis 2a: Parents reporting less effective coping behaviors were hypothesized to have children with poorer outcomes at follow-up.
CHAPTER 2
METHOD

Participants

Participants were 25 youth-parent dyads diagnosed with CF at the University of Florida and Virginia Commonwealth University who were part of a larger study that assessed sleep and physical activity levels. Inclusion criteria for the current study were: (1) children were between 4-12-years-old at the time of study entry and have at least one parent or legal guardian; (2) children had a documented current diagnosis of cystic fibrosis verified by the child’s electronic medical record; (3) child and parent were fluent in English; and (4) children lived with the parent or legal guardian participating in the study. Exclusion criteria were: (1) child had history of a lung transplant; (2) child had been hospitalized within the past two weeks; and (3) child had experienced an exacerbation, defined as requiring therapy with oral or IV antibiotics in the past two weeks. These exclusion criteria were selected to account for increases in parent reported mental health symptoms and changing child health outcomes during hospitalization and exacerbation periods (Quittner et al., 2008; Quittner et al., 2012). If participants were excluded due to previous hospitalizations or exacerbations, they were invited to participate at subsequent appointments. Please see Figure 2-1 for detailed study flow information.

Descriptive statistics for baseline and follow-up are displayed in Table 3-1. Children were an average of 8.1 years of age ($SD = 2.6$). A majority of the children in the sample were Caucasian (76.9%), while 19% identified as a racial/ethnic minority. Median weight at baseline was 28.76 kg ($SD = 10.01$), height was 1.27 m ($SD = .14$), and BMI was 17.32 ($SD = 2.77$). Median number of hospitalizations in the past year was
1.00 and ranged from 0 - 4.00 ($M = .96$). The range from baseline appointment to follow-up appointment was 5.22 - 11.08 months with a median number of 7.4 months ($M = 7.32$). Variation in time from baseline to follow-up data collection was due to delays stemming from exacerbations or hospitalizations. The majority of children were diagnosed with cystic fibrosis mutation Delta F508 homozygous only (54%) with additional mutations including combinations of Delta F508 and additional mutations (31%), and other (12%). Please see additional mutations in Table 3-1. Baseline time point FEV1 % predicted ranged from 63% -120% ($M = 96.36$, $SD = 13.53$) with follow-up lung function ranging from 84% - 123% ($M = 98.00$, $SD = 12.55$).

**Procedure**

Interested families were screened in the pediatric cystic fibrosis clinics at the University of Florida or Virginia Commonwealth University to determine if they met inclusion criteria. Dyads who expressed interest in participating and met eligibility criteria completed informed consent in the CF clinics. Written consent and assent were obtained from all participants, with the exception of children under the age of six who were not required to provide written assent. After completing informed consent, participants were emailed a website link that directed them to the study questionnaires on REDCap, a secure, web-based database. Participants entered their unique study identification and received instructions to complete each questionnaire before proceeding to the next questionnaire. Participants received scripted phone calls and emails to prompt them to log in to complete measures. For participants without access to a personal computer or a smart device, a paper and pencil copy of study questionnaires and prepaid envelopes to return them in were provided. Six months following the completion of baseline measures, participants received an email prompting
them to complete the depression, anxiety, and HRQOL online questionnaires through REDCap. Some participants were delayed due to exacerbations or hospitalizations. Concurrently, research personnel contacted participants on three consecutive days to complete an additional 24-hour recall to monitor treatment adherence (please see below for additional adherence information). Participants were compensated $10.00 following completion of questionnaires at baseline and follow-up and $20.00 for completion of the phone recall measure at baseline and at the six-month follow-up time point.

**Measures**

Parents completed the following questionnaires (see Appendices A and B).

**Demographic Questionnaire**

Parents completed basic demographic information, including the child’s date of birth, sex, race, ethnicity, and child’s year in school. They also completed questions on their marital status, education level, ethnicity, and occupation as well as their partner’s ethnicity, education level, and occupation.

**Medical chart review**

Medical information was abstracted from the electronic medical record at their baseline clinic appointment as well as at their six-month follow-up appointment. All charts were abstracted by research personnel and verified by a member of the medical staff. Specific extraction information included the child’s height, weight, CF mutation classification, medication regimen, airway clearance regimen, history of CF-related hospitalization and exacerbations, and lung function at their most recent clinic visit [e.g., forced expiratory volume in one second (FEV1), and forced expiratory volume in one second predicted (FEV1 % predicted) forced vital capacity (FVC), forced expiratory flow (FEF), total lung capacity (TLC)]. FEV1 % predicted was used as the primary outcome
for lung function in the current study due to its frequent use in the pediatric CF literature (Quittner et al., 2014). Baseline medical chart review assessed information from the previous year, while follow-up medical chart review assessed the 6-month time window since study entry.

**Parent Depressive Symptoms**

Parents completed the nine-item Patient Health Questionnaire depression scale (PHQ-9; Spitzer et al., 1996). Respondents reported on how often they were bothered by nine different problems during the last two weeks (e.g., trouble falling or staying asleep, or sleeping too much). Response options include “not at all,” “several days,” “more than half the days,” or “nearly every day,” scored as 0, 1, 2, and 3, respectively. This measure has been well-validated in community and medical populations (Martin, Rief, Klaiberg & Braehler, 2006). Scores were summed to yield a total score. Commonly used clinical cut-offs are as follows: values of 1-4 indicate minimal depression, 5-9 indicates mild depression, 10-14 indicates moderate depression, 15-19 indicates moderately severe depression, and 20-27 severe depression (Spitzer et al., 1996). This measure is a reliable and valid assessment of depression severity and was recommended by the TIDES study as a symptom screener in parents of children with CF (Quittner et al., 2014; Kroenke, Spitzer, & Williams, 2001). Cronbach’s alpha for the current study was .83, indicating good internal reliability.

**Parent Anxiety Symptoms**

Parents completed the Generalized Anxiety Disorder Screener (GAD-7; Spitzer, Kroenke, Williams, & Lowe, 2006). Parents were asked to report on how often they have been bothered by seven different problems during the last two weeks (e.g., not being able to stop or control worrying). Response options are “not at all,” “several days,”
“more than half the days,” and “nearly every day,” scored as 0, 1, 2, and 3, respectively. The sum of these scores was calculated, yielding a total score. Total scores can range from 0 to 21. Established clinical cut-offs are: ≥ 5 represents mild anxiety symptoms, ≥ 10 representing moderate anxiety symptoms, and ≥ 15 represents severe anxiety (Spitzer, Kroenke, Williams, & Lowe, 2006). The GAD-7 has demonstrated good reliability and validity in measuring anxiety and was recommended by the TIDES study as a symptom screener in parents of children with CF (Quittner et al., 2014; Lowe et al., 2008). Cronbach’s alpha for the current study was .95, indicating excellent internal reliability.

**Role-play Inventory of Situations and Coping Skills (RISCS) Vignettes**

The RISCS (DiGirolamo et al., 1997) contains twenty-nine context-specific vignettes in eleven domains designed to elicit parental coping behaviors in a variety of common challenging situations that parents of children with CF encounter as well as a total coping effectiveness score. Specific domains include treatment adherence, parent-child relationship, eating/weight gain, medical procedures and symptoms, growing up/health issues, school issues, sibling issues, witnessed child peer issues, parent finance issues, and parent partner issues. The total coping effectiveness score used in the current study is calculated from the average of all completed vignettes. Parents provided ratings for two questions related to each independent vignette. First, they rated the vignette based on how frequently the situation occurred over the past year (1 = not at all frequent; 5 = a lot). Second, the parent rated the difficulty of the situation based on a similar scale (1 = not at all difficult; 5 = extremely difficult). Following these ratings, participants produced a coping response for each vignette. Following published guidelines (DiGirolamo et al., 1997), coping responses were coded for effectiveness.
using manualized criteria on a 4-point scale ranging from an extremely incompetent/ineffective (e.g., “1” = “We’d just skip all of the treatment because it’s very important that he get to do things with other kids”) to an extremely competent/effective response (e.g., “4” = “We explain how important the treatment is so that she knows she has to do it. She would have to leave her friend’s house a little bit earlier so that the treatment was done before bedtime”).

Trained research personnel rated coping responses using manualized guidelines (DiGirolamo et al., 1997). Responses were rated as more effective if they decreased the frequency or perceived difficulty of the problem or reduced long-term negative consequences (DiGirolamo et al., 1997). A trained staff member coded all responses for each participant. Two additional trained raters independently scored a random subset (25%) of the participants completing the RISCS. Cohen's κ was run to determine if there was agreement between three raters’ judgment on the 29 coping specific scenarios in the RISCS. There was substantial agreement κ = .66, p < .001 between rater 1 and 2; κ = .78, p < .001 between raters 1 and 3, and κ = .83, p < .001 between rater 2 and 3. Average κ = .76 which exceeds acceptability (Landis & Koch, 1977).

**Child Health-Related Quality of Life**

Parents completed the Cystic Fibrosis Questionnaire-Revised parent version (CFQ-R; Modi & Quittner, 2003; Quittner, Buu, Messer, Modi, & Watrous, 2005). This is a 44-item questionnaire for parents of children ages 6-13-years-old covering eleven domains of HRQOL on 4-point scales (“a lot of difficulty,” “some difficulty,” “a little difficulty,” “no difficulty;” “always,” “often,” “sometimes,” “never;” “very true,” “somewhat true,” “somewhat false,” “very false;” ”a great deal,” “often,” “sometimes,” “never”). Specifically, it covers four broad domains and seven CF symptom-specific domains.
Broad domains include 1) physical symptoms; 2) vitality; 3) emotional functioning; and 4) school functioning. Specific domains assess challenges related to mealtimes, the degree of treatment interference with daily activities, children’s comparisons to peers, challenges associated with weight gain, the degree of respiratory distress (e.g., wheezing, mucus appearance, etc.), and gastrointestinal distress (e.g., diarrhea, pain, etc.).

Given the focus of current study assessing relation between parent-reported symptoms of depression, anxiety, and coping behavior on child health outcomes, the physical symptoms, vitality, and emotional functioning domains were used in analysis. School functioning was not used due to high number of homeschooled participants or participants too young for school ($n = 7; 27\%$). Specifically, this domain asks parents to rate their child’s health (e.g., “excellent,” “good,” “fair,” or “poor”) as well as rating the degree to which their child is able to feel healthy and to live a “normal” life. The CFQ-R is a validated HRQOL measure for CF (Modi & Quittner, 2003; Quittner, Buu, Messer, Modi, & Watrous, 2005). Raw scores were converted into standardized scores for each of the 4 scales with higher scores representative of better HRQOL. Cronbach’s alpha for the current study was .91, indicating excellent internal reliability.

**Adherence**

Parents completed the Daily Phone Diary (DPD), a validated and well-studied computer-based interview, which assesses daily activities and mood states over a 24-hour period on three consecutive days (2 weekdays, 1 day of the weekend) at baseline assessment and 6-month follow-up (Quittner & Espelage, 1999; Modi & Quittner, 2006). This information was collected by asking parents to recall all events their child engaged in over the past 24 hours based on five-minute increments. The DPD assessed
activities, social interactions, emotions, and time engaged in each event. For each event lasting longer than 5 minutes, parents described the duration, companions, and their child’s mood. This measure is commonly used in CF to assess adherence to medical regimens including airway clearance treatments, dietary recommendations, and medications taken over the course of the day. While diaries are a form of self-report, the DPD reduces social desirability responding due to eliciting information regarding all activities lasting longer than five minutes (Modi, Lim, Yu, Geller, Wagner, & Quittner, 2006). Adherence to airway clearance treatments was used as the primary adherence variable of interest. Airway clearance adherence is comprised of the ratio of reported treatments completed (e.g., vest treatments) compared to prescribed treatment regimen based on medical record review.

\[
Adherence (\%) = \frac{\text{# of Completed Airway Clearance Treatments}}{\text{# of Prescribed Airway Clearance Treatments}} \times 100
\]

Phone calls typically lasted 10-15 minutes, with prompts such as, “After you finished dinner at 7:00 PM, what did you do next?” Responses were automatically computer-coded in accordance with activity codes such as self-care (e.g., getting ready for bed, napping, etc.), medical care (e.g., pulmozyme, visit to pulmonologist, etc.), household tasks (e.g., cleaning, yard work etc.), recreation (e.g., playing outside, playing a sport), and school (e.g., getting to or from school, eating a meal at school). The DPD has demonstrated validity in CF populations (Modi & Quittner, 2006) and is a “well-established” measure of adherence (Quittner et al., 2008).

**Statistical Analyses**

Descriptive statistics were used to report demographics, medical characteristics, and outcome variables within the current sample. Preliminary analyses were conducted
using either Pearson product-moment correlations or t-tests to examine the relationship between demographic variables (e.g., age, ethnicity) and outcomes of interest. Variables with significant associations with outcome variables of interest were included as covariates during hypothesis testing.

Separate linear regression models were used to examine whether parent mental health at baseline was associated with child adherence, lung function, and HRQOL. Applicable covariates were entered into Block 1 and predictor variables of interest were entered in Block 2. Similarly, to examine whether the effectiveness of parent coping behaviors at baseline was associated with child adherence, lung function, and HRQOL, separate linear regression models were used. Applicable covariates were entered into Block 1 and predictor variables of interest were entered into Block 2.

To compare child health outcomes (e.g., adherence, lung function, and HRQOL), ANOVA or ANCOVA analyses were conducted to assess group differences between parents reporting symptoms of depression or anxiety above and below mild clinical elevations. The same analyses were conducted using the moderate clinical cut-off scores.

Separate mediational analysis using hierarchical linear relationships were proposed to determine if effectiveness of parent-reported coping behavior mediates the relationship between parent-reported symptoms of depression and anxiety on child health outcomes. Mediation analyses were proposed in accordance with the approach outlined by Baron & Kenny (1986). This method provides a systematic approach to mediation by running a series of regression models estimating (1) the total effect of the independent variables on the dependent variables (c-path; PHQ-9 and GAD-7 on child
health outcome), (2) the effect of the independent variables on the mediator (a-path; PHQ-9 and GAD-7 on effectiveness of parent-reported coping behavior), (3) the effect of the mediator on the dependent variable (b-path; parent-reported coping behavior on child health outcome), and the direct effect of the independent variable on the dependent variable when accounting for the mediators (c’-path). Mediation analyses will only be conducted in the context of a previously established association between parent-reported symptoms of depression or anxiety and a child health outcome in Aim 1.

It was initially proposed to use separate linear regression models to examine the relation between baseline parent-reported symptoms of depression, anxiety, and effective coping behaviors on six-month follow-up treatment adherence (DPD), lung function, and child HRQOL. However, given the modest sample size at the follow-up time point (n = 10) and the exploratory nature of this aim, correlational analyses were conducted.

Cohen’s $f^2$ was calculated as a measure of effect size for all regression analyses. Standard conventions for $f^2$ are as follows: .02 is considered small, .15 is considered medium, and .35 is considered large (Cohen, 1988). Cohen’s $d$ was calculated as a measure of effect size for all ANOVA and ANCOVA analyses. Standard conventions for $d$ are as follows: .2 is considered small, .5 is considered medium, and .8 is considered large (Cohen, 1988). Analyses were conducted using IBM SPSS, Version 24.
CHAPTER 3
RESULTS

Preliminary Analyses

Statistically significant relationships were found between child’s age and the adherence score \( r = -.511, p = .015 \) and the CFQ-R emotional functioning subscale \( r = -.665, p = .002 \). Age was subsequently included as a covariate in applicable models.

Characterizing Parent Mental Health

Descriptive information related to scores on the PHQ-9 and GAD-7 is presented in Table 1.2. Slightly less than one-third of the sample endorsed clinically elevated (i.e., mild or higher symptoms) levels of depression at baseline \((n = 5; 19\%)\). For anxiety symptoms, just under half the parents endorsed clinically elevated levels at baseline \((n = 11, 42.1\%)\). At follow-up, a similar percentage of the sample endorsed clinically elevated symptoms of depression \((n = 3, 30\%)\) as well as a similar percentage of clinically elevated symptoms of anxiety \((n = 5, 50\%)\).

Association of Parent Mental Health with Adherence

Age was included as a covariate in all adherence analyses. When assessing parent-reported symptoms of depression, results revealed that the overall model predicting total treatment adherence scores was not statistically significant, \( F(2, 17) = 3.15, p = .069, R^2 = .270, \hat{f} = .37 \). Parent-reported symptoms of depression \((\beta = .04, p = .864; \hat{f} = .001)\) were not related to child treatment adherence. Additionally, when assessing parent-reported symptoms of anxiety, analyses revealed that the overall model predicting total adherence was not statistically significant, \( F(2, 18) = 3.40, p = .056, R^2 = .274, \hat{f} = .38 \). Parent-reported symptoms of anxiety \((\beta = -.162, p = .861; \hat{f} = .001)\) did not account for a significant amount of variance in total adherence scores.
Association of Parent Mental Health with Lung Function

Results revealed that the overall model predicting lung function (i.e., FEV1 % predicted), $F(1, 18) = .012, p = .916, R^2 = .001, \hat{f}^2 = .001$, was not statistically significant. Parent-reported symptoms of depression ($\beta = -.025, p = .916$) did not account for a significant amount of variance in lung function scores. Additionally, when assessing parent-reported symptoms of anxiety, analyses revealed that the overall model predicting lung function was not statistically significant, $F(1, 20) = .103, p = .752, R^2 = .005, \hat{f}^2 = .005$. Parent-reported symptoms of anxiety ($\beta = -.071, p = .752$) did not account for a significant amount of variance in lung function scores.

Association of Parent Mental Health with Child Health-Related Quality of Life

Physical Symptoms

When assessing parent-reported symptoms of depression, results revealed the overall model predicting child CFQ-R physical symptoms was not significant, $F(1, 19) = .077, p = .784, R^2 = .004, \hat{f}^2 = .004$. Parent-reported symptoms of depression ($\beta = .064, p = .784$) did not account for a significant amount of variance in CFQ-R physical symptoms domain. Similarly, results assessing symptoms of anxiety revealed the overall model predicting HRQOL physical symptoms was not significant, $F(1, 21) = .254, p = .619, R^2 = .012, \hat{f}^2 = .012$. Parent-reported symptoms of anxiety ($\beta = .109, p = .619$) did not account for a significant amount of variance in physical symptoms.

Emotional Functioning

When assessing symptoms of depression, results revealed the overall model predicting child CFQ-R emotional functioning was not significant, $F(2, 18) = 2.944, p = .078, R^2 = .246, \hat{f}^2 = .326$. Parent-reported symptoms of depression ($\beta = -.090, p = .681; \hat{f}^2 = .007$) did not account for a significant amount of variance in CFQ-R emotional
functioning domain beyond child age. Additionally, when assessing parent-reported symptoms of anxiety, analyses revealed that the overall model predicting child HRQOL emotional functioning domain was significant, $F(2, 20) = 2.514, p = .106, R^2 = .201, \hat{\eta}^2 = .252$. Parent-reported symptoms of anxiety ($\beta = .073, p = .753; \hat{\eta}^2 = .005$) did not account for a significant amount of variance in emotional functioning beyond age.

**Vitality**

When assessing symptoms of depression, results revealed the overall model predicting child CFQ-R vitality was not significant, $F(1, 19) = .03, p = .865, R^2 = .002, \hat{\eta}^2 = .002$. Parent-reported symptoms of depression ($\beta = .039, p = .865$) did not account for significant variance in CFQ-R vitality scores. Additionally, results assessing symptoms of anxiety revealed the overall model predicting HRQOL vitality domain was not significant, $F(1, 21) = .001, p = .982, R^2 = .000, \hat{\eta}^2 = .000$. Parent-reported symptoms of anxiety ($\beta = -.005, p = .982$) did not account for a significant amount of variance in vitality.

**Association of Parent Coping Behavior with Treatment Adherence**

Results revealed the overall model predicting treatment adherence was not significant, $F(2, 19) = 3.78, p = .666, R^2 = .268, \hat{\eta}^2 = .37$. Parent-reported coping behaviors ($\beta = .089, p = .666; \hat{\eta}^2 = .24$) did not account for a significant amount of variance in adherence beyond child age.

**Association of Parent Coping Behavior with Lung Function**

The overall model predicting lung function was significant, $F(1, 22) = 8.07, p = .01, R^2 = .268, \hat{\eta}^2 = .367$. Parent-reported coping behaviors ($\beta = .518, p = .01$) accounted for significant variance in lung function. Therefore, higher levels of parent coping effectiveness were associated with increased lung function.
Association of Parent Coping Behavior with Child Health-Related Quality of Life

Physical Symptoms

Results revealed the overall model predicting HRQOL physical symptoms was significant, $F(1, 22) = 10.533, p = .004$, $R^2 = .324$, $f^2 = .479$. Parent-reported coping behavior ($\beta = -.569$, $p = .004$) accounted for a significant amount of variance.

Emotional Functioning

Results revealed the overall model predicting HRQOL emotional functioning was not significant, $F(2, 21) = 2.725, p = .089$, $R^2 = .206$, $f^2 = .259$. Effectiveness of parent-reported coping behaviors ($\beta = -.114$, $p = .576$; $f^2 = .012$) did not account for a significant amount of variance in emotional functioning after controlling for child age.

Vitality

Results revealed the overall model predicting HRQOL vitality was not significant, $F(1, 22) = 1.923, p = .179$, $R^2 = .080$, $f^2 = .087$. Effectiveness of parent-reported coping behaviors ($\beta = -.284$, $p = .179$) did not account for a significant amount of variance in vitality.

Comparisons of Outcomes between Parents Above versus Below Clinical Cut-offs for Depression

Treatment Adherence

ANCOVA analyses controlling for child age revealed that treatment adherence was not significantly different, $F(1, 17) = 1.002$, $p = .331$, $d = .49$, among children whose parents were above ($M = 59.80$, $SD = 48.13$) or below ($M = 86.53$, $SD = 23.01$) the mild clinical cut-off for depressive symptoms. ANCOVA analyses controlling for child age revealed that treatment adherence was not significantly different, $F(1, 17) = 2.23$, $p$
=.154, d = .72, among children whose parents were above (M = 94.33, SD = 9.81) or
below (M = 77.29, SD= 33.94) the moderate clinical cut-off for depressive symptoms.

Lung Function

ANOVA analyses that lung function was not significantly different, F(1, 21) = 
2.55, p =.125, d = .7, among children whose parents were above (M = 100.5, SD =
11.36) or below (M = 91.8, SD= 12.94) the mild clinical cut-off for depressive symptoms. 
ANOVA analyses controlling for child age revealed that lung function was not
significantly different, F(1, 21) = 1.623, p =.217, d = .56, among children whose parents 
were above (M = 100.5, SD = 12.03) or below (M = 92.82, SD= 12.89) the moderate 
clinical cut-off for depressive symptoms.

Health-Related Quality of Life

Physical Symptoms: ANOVA analyses revealed that CFQ-R physical symptoms 
was not significantly different, F(1, 22) = 1.599, p =.219, d = .67, among children whose 
parents were above (M = 94.27, SD = 7.02) or below (M = 83.71, SD= 20.45) the mild 
clinical cut-off for depressive symptoms. ANOVA analyses revealed that CFQ-R 
physical symptoms was not significantly different, F(1, 22) = 2.605, p =.121, d = .69, 
among children whose parents were above (M = 97.22, SD = 5.05) or below (M = 85.88, 
SD= 16.74) the moderate clinical cut-off for depressive symptoms.

Emotional Functioning: ANCOVA analyses controlling for child age revealed
that CFQ-R emotional functioning was not significantly different, F(1, 18) = .394, p
=.538, d = .54, among children whose parents were above (M = 94.27, SD = 7.02) or
below (M = 85.94, SD= 17.8) the mild clinical cut-off for depressive symptoms.

ANOVA analyses controlling for child age revealed that CFQ-R emotional functioning
was not significantly different, $F(1, 21) = .752, p = .396, d = .38$, among children whose parents were above ($M = 85.56, SD = 12.94$) or below ($M = 81.85, SD = 17.39$) the moderate cut-off for depressive symptoms.

**Vitality:** ANOVA analyses revealed that CFQ-R vitality was not significantly different, $F(1, 22) = .039, p = .846, d = .09$, among children whose parents were above ($M = 73.33, SD = 18.52$) or below ($M = 74.58, SD = 12.47$) the mild clinical cut-off for depressive symptoms. ANOVA analyses revealed that CFQ-R vitality was not significantly different, $F(1, 22) = .495, p = .489, d = .30$, among children whose parents were above ($M = 77.78, SD = 10.04$) or below ($M = 72.93, SD = 15.59$) the moderate clinical cut-off for depressive symptoms.

**Comparisons of Outcomes between Parents Above versus Below Clinical Cut-offs for Anxiety**

**Treatment Adherence**

ANOVA analyses controlling for child age revealed that adherence was not significantly different, $F(1, 18) = .141, p = .712, d = .18$, among children whose parents were above ($M = 71.50, SD = 32.62$) or below ($M = 86.27, SD = 29.65$) the mild clinical cut-off for symptoms of anxiety. ANCOVA analyses controlling for child age revealed that adherence was not significantly different, $F(1, 18) = .165, p = .69, d = .19$, among children whose parents were above ($M = 80.50, SD = 26.78$) or below ($M = 78.73, SD = 33.72$) the moderate clinical cut-off for symptoms of anxiety.

**Lung Function**

ANOVA analyses revealed that lung function was not significantly different, $F(1, 20) = .772, p = .39, d = .39$, among children whose parents were above ($M = 96.4, SD = 15$) or below ($M = 91.75, SD = 9.69$) the mild clinical cut-off for symptoms of anxiety.
ANOVA analyses revealed that lung function was not significantly different, $F(1, 20) = .501, p = .487, d = .31$, among children whose parents were above ($M = 90.4, SD = 16.61$) or below ($M = 94.88, SD = 11.16$) the moderate clinical cut-off for symptoms of anxiety.

**Health-Related Quality of Life**

**Physical Symptoms**: ANOVA analyses revealed that CFQ-R physical symptoms was not significantly different, $(1, 21) = .014, p = .907, d = .06$, among children whose parents were above ($M = 88.64, SD = 16.68$) or below ($M = 87.85, SD = 15.23$) the mild clinical cut-off for symptoms of anxiety. ANOVA analyses revealed that CFQ-R physical symptoms was not significantly different, $F(1, 21) = .144, p = .708, d = .17$, among children whose parents were above ($M = 86.11, SD = 22.15$) or below ($M = 88.97, SD = 13.33$) the moderate clinical cut-off for symptoms of anxiety.

**Emotional Functioning**: ANCOVA analyses controlling for child age revealed that CFQ-R emotional functioning was not significantly different, $F(1, 20) = .235, p = .633, d = .22$, among children whose parents were above ($M = 77.58, SD = 15.31$) or below ($M = 86.67, SD = 15.31$) the mild clinical cut-off for symptoms of anxiety. ANCOVA analyses controlling for child age revealed that CFQ-R emotional functioning was not significantly different, $F(1, 20) = .018, p = .894, d = .06$, among children whose parents were above ($M = 82.22, SD = 15.01$) or below ($M = 82.35, SD = 17.31$) the moderate clinical cut-off for symptoms of anxiety.

**Vitality**: ANOVA analyses revealed that CFQ-R vitality was not significantly different, $F(1, 21) = 1.29, p = .269, d = .50$, among children whose parents were above ($M = 70.91, SD = 16.40$) or below ($M = 77.78, SD = 12.50$) the mild clinical cut-off for
symptoms of anxiety. ANOVA analyses revealed that CFQ-R vitality was not significantly different, $F(1, 21) = .728, p = .403, d = .38$, among children whose parents were above ($M = 78.89, SD = 7.79$) or below ($M = 72.94, SD = 16.24$) the moderate clinical cut-off for symptoms of anxiety.

**Mediational Analyses of Coping Behavior on Parent Mental Health**

Parent reported symptoms of depression and anxiety did not significantly predict child health outcomes. Therefore, due to null findings for the main effects, mediational analyses to determine if effectiveness of parent-reported coping behavior mediates the relationship between parent-reported symptoms of depression and anxiety on child health outcomes using hierarchical linear regression were not conducted.

**Exploratory Analyses: Associations of Parent Mental Health and Coping Behaviors at Baseline with Child Health Outcomes at Follow-up**

Parent-reported symptoms of depression at baseline were not significantly correlated with any child health outcomes at follow-up (adherence: $r(8) = .345, p = .363$; lung function: $r(8) = -.387, p = .239$; CFQ-R physical symptoms: $r(8) = -.577, p = .134$; CFQ-R emotional functioning: $r(8) = -.49, p = .217$; CFQ-R vitality: $r(8) = -.046, p = .923$.

Parent-reported symptoms of anxiety at baseline were not significantly correlated with any child health outcomes at follow-up (adherence: $r(8) = -.047, p = .904$; lung function: $r(8) = -.142, p = .677$; CFQ-R physical symptoms: $r(8) = -.564, p = .146$; CFQ-R emotional functioning: $r(8) = -.526, p = .181$; CFQ-R vitality: $r(8) = .186, p = .69$.

Parent-reported effectiveness of coping behaviors (RISCS total score) were not significantly related to any child health outcomes at follow-up (adherence: $r(8) = -.453, p = .188$; lung function: $r(8) = .206, p = .544$; CFQ-R physical symptoms: $r(8) = -.062, p = .
.883; CFQ-R emotional functioning: \( r(8) = -.143, p = .736 \); CFQ-R vitality: \( r(8) = .613, p = .143 \). Please see Table 3-6 for additional details.
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<td>Table 3-2. Parent Characteristics with Depression and Anxiety Cut-offs</td>
<td>BASELINE</td>
<td>FOLLOW-UP</td>
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<tr>
<th>Table 3-3. Regression Models with Depression (PHQ-9) as the Predictor</th>
<th>Outcome Variable</th>
<th>$R^2$</th>
<th>$\Delta R^2$</th>
<th>$B$</th>
<th>Sig.</th>
<th>Overall $f^2$</th>
<th>$\bar{f}$</th>
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<tbody>
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<td>1. Adherence$^a$</td>
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<td>.001</td>
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<td>.001.</td>
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<td>-.090</td>
<td>.078</td>
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<td>.007</td>
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<td>.865</td>
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* $p<.10$; ** $p<.05$; $a$ = Separate model controlling for age.

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<th>Table 3-4. Regression Models with Anxiety (GAD-7) as the Predictor</th>
<th>Outcome Variable</th>
<th>$R^2$</th>
<th>$\Delta R^2$</th>
<th>$B$</th>
<th>Sig.</th>
<th>Overall $f^2$</th>
<th>$\bar{f}$</th>
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<td>.001</td>
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<tr>
<td>Physical</td>
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<td>.064</td>
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<td>.000</td>
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*p<.10; **p<.05; $a$ = Separate model controlling for age.
Table 3-5. Regression Models with Coping Behavior (RISCs) as the Predictor

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<tr>
<th>Outcome Variable</th>
<th>$R^2$</th>
<th>$\Delta R^2$</th>
<th>$\beta$</th>
<th>Sig.</th>
<th>Overall $f^2$</th>
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<td>.367</td>
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<td>.004**</td>
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$^a$p<.10; **p<.05; a = Separate model controlling for age.

Table 3-6. Correlations between Baseline Parent-Reported Symptoms of Depression, Anxiety, and Coping Behavior and Follow-Up Child Outcomes

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<th>Coping</th>
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<td>3. CFQ-R: Physical Symptoms</td>
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<td>-.062</td>
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<tr>
<td>4. CFQ-R: Emotional Functioning</td>
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<td>-.526</td>
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<tr>
<td>5. CFQ-R: Vitality</td>
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<td>.613</td>
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</tbody>
</table>

*p < .05. **p < .01
Participants consented  
\((n = 32)\)

Completed Baseline Measures  
DPD \((n = 23)\)  
FEV 1 % \((n = 25)\)  
PHQ \((n = 21)\)  
GAD \((n = 23)\)  
RISCS \((n = 25)\)  
CFQ-R \((n = 25)\)

Withdrawn (Death; moved)  
\((n = 2)\)

Completed Follow-up Measures  
DPD \((n = 10)\)  
FEV 1 % \((n = 10)\)  
PHQ \((n = 10)\)  
GAD \((n = 10)\)  
CFQ-R Physical \((n = 10)\)  
CFQ-R Emotional \((n = 10)\)  
CFQ-R Vitality \((n = 9)\)
Figure 3-2. Means of adherence in mild versus moderate depression groups

Cohen’s $d$ (mild) = .49; Cohen’s $d$ (moderate) = .72

Figure 3-3. Means of adherence in mild versus moderate anxiety groups

Cohen’s $d$ (mild) = .18; Cohen’s $d$ (moderate) = .19
Figure 3-4. Means of lung function in mild versus moderate depression groups

Cohen’s $d$ (mild) = .7; Cohen’s $d$ (moderate) = .56

Figure 3-5. Means of lung function in mild versus moderate anxiety groups

Cohen’s $d$ (mild) = .39; Cohen’s $d$ (moderate) = .31
Figure 3-6. Means of HRQOL in mild depression group

Cohen’s $d$ (Physical) = .67
Cohen’s $d$ (Emotional) = .54
Cohen’s $d$ (Vitality) = .09

Figure 3-7. Means of HRQOL in moderate depression group

Cohen’s $d$ (Physical) = .69
Cohen’s $d$ (Emotional) = .38
Cohen’s $d$ (Vitality) = .30
Figure 3-8. Means of HRQOL in mild anxiety group

Cohen’s \( d \) (Physical) = .06
Cohen’s \( d \) (Emotional) = .22
Cohen’s \( d \) (Vitality) = .50

Figure 3-9. Means of HRQOL in moderate anxiety group

Cohen’s \( d \) (Physical) = .17
Cohen’s \( d \) (Emotional) = .06
Cohen’s \( d \) (Vitality) = .38
CHAPTER 4
DISCUSSION

The overarching aim of this study was to extend the extant research assessing how parent reported symptoms of depression, anxiety, and coping behaviors were related to child health outcomes (e.g., adherence, lung function, and health-related quality of life) in 4-12 year-olds with CF. This study sought to address prior methodological gaps in the existing pediatric CF literature on the impact of parental symptoms of psychological distress and effectiveness of coping behaviors on child health outcomes by utilizing objective medical outcomes (e.g., lung function), an empirically supported measure of CF regimen adherence, and a CF-specific measure of HRQOL.

Pediatric psychology continues to incorporate Bronfenbrenner’s ecological systems theory into research and clinical practice due to the significance of parental behaviors and functioning on child outcomes within the family system (Kazak, Simms, & Rourke, 2002). Results from the current study contribute to the growing literature recognizing the important impact of parent functioning on child health outcomes and HRQOL in chronic illness populations. Additionally, this is one of the few studies assessing the implications of the effectiveness of parent generated coping behaviors on child health outcomes in CF. By focusing on objective child health outcomes related to parental psychological distress and coping behavior this study addressed prior research gaps, extends current research, and provides implications for future clinical practice.

Parent-reported Symptoms of Depression and Anxiety

Rates of parent-reported symptoms of anxiety and depression in this study are comparable to previous studies, including The International Depression Epidemiological
Study (TIDES) (Besier et al., 2011; Driscoll, Montag-Leifling, Acton, & Modi, 2009; Quittner et al., 2014). Overall rates of parent-reported symptoms of depression in the CF literature range between 20-35% (Havermans, Colpaert, & Dupont, 2008; Quittner et al., 2016) while symptoms of anxiety range between 17-48% (Besier et al., 2011; Quittner et al., 2014). Findings from the current study fit within these ranges of parent-reported psychological distress. Specifically, 19% \( (n = 5) \) of parents reported at least mild elevations in depression at baseline and 30% \( (n = 3) \) of parents reported at least mild elevations at follow-up. Additionally, the current study found that 42% \( (n = 11) \) of parents reported at least mild elevations in symptoms of anxiety at baseline while 50% \( (n = 5) \) reported at least mild elevations at follow-up. Collectively, these results indicate that a meaningful subset of parents of children with CF experience psychological distress. This further underscores the importance of ongoing efforts within the pediatric CF community to identify and provide mental health services for parents of children who are distressed at pediatric CF clinic visits (Quittner et al., 2016).

**Association of Parent Mental Health and Coping with Lung Function and Adherence**

Counter to hypotheses, parent-reported symptoms of anxiety and depression were not related to their child’s treatment adherence, lung function, or HRQOL at baseline or follow-up. Findings from the current study are in contrast to previous work indicating a robust relationship between poorer parent functioning and child pulmonary functioning, distress and internalizing symptoms, and quality of life (Fedele et al., 2013; Patterson, McCubbin, & Warwick, 1990; Quittner et al., 1996; Quittner, Drotar, & Ievers, 1998; Ryan et al., 2010). Specific to CF, prior research has also found an association between elevated parent and child distress ratings, higher maternal depression
symptoms related to poorer medication adherence, and higher overall parent-reported symptoms of depression associated with lower enzyme adherence and lack of weight gain (Besier & Goldbleck, 2011; DeLambo et al., 2004; Everhart et al., 2014; Ryan et al., 2010; Smith, Modi, Quittner, & Wood, 2010; Quittner, Barker, & Gellar, 2007; Quittner et al., 2014).

Lack of an association between parent-reported symptoms of depression and anxiety with their child’s CF airway clearance adherence in the current study is inconsistent with previous literature linking higher parental psychological distress with lower adherence rates (Quittner, Barker, Gellar, Butt, & Gondor, 2007). While the association is not significant, levels of airway clearance adherence in the current sample ($M_{\text{baseline}} = 51.43\%$, $SD_{\text{baseline}} = 28.18$; $M_{\text{follow-up}} = 52.13\%$, $SD_{\text{follow-up}} = 26.68$) are largely commensurate with the extant literature (DeLambo, levers-Landis, Drotar, & Quittner, 2004; Eakin & Riekert, 2013; levers et al., 1999; Modi et al., 2006; Zindani, Streetman, & Nasr, 2006; Modi, Lim, Yu, Gellar, Wagner, & Quittner, 2006; DiGirolamo, Quittner, Ackerman, & Stevens, 1997; Llorente, Garcia, & Martin, 2008). While it was hypothesized that higher parent-reported psychological distress would predict lower treatment adherence, it is possible the association was not found due to the young age of this sample. Adherence for this age-range is nearly exclusively dependent upon parent treatment implementation. Given these expectations, parents may view child treatment completion as a higher priority than managing personal symptoms of psychological distress. This is anecdotally supported with responses on the RISCS measure where parents reported treatment completion as their main priority, regardless of personal desires (e.g., sacrificing opportunities for self-care to ensure treatment
completion; “I have to put all my energy and time into my children so I have no time for myself”).

Lack of an association between parental psychological distress and lung function could potentially relate to the health status of our sample at the time of assessment. According to pulmonary function test results, FEV1 % predicted values suggest that the majority of the children in this sample were classified as mild illness severity at the time of measurement (FEV1 % predicted ≥ 70; $M_{\text{baseline}} = 96.36\%$, $SD_{\text{baseline}} = 13.53$; $M_{\text{follow-up}} = 98\%$, $SD_{\text{follow-up}} = 12.55$) based on established cut-offs (Taussig, 1995). Although speculative, findings from the current study could stem from the study’s inclusion criteria surrounding pulmonary exacerbations. Since families were excluded from participation during CF exacerbations, it is possible that the collected lung function values are somewhat inflated or reflective of a recent medical intervention (e.g., hospitalization, IV antibiotics). Given the established link between disease severity and parent-reported distress (Quittner et al., 2014), it is possible that a link between parent distress and lung function could be present in the context of a child experiencing increased CF symptoms which is not captured in the context of the current study. Therefore, assessing parents’ symptoms of distress during exacerbations and hospitalizations appears prudent.

Alternatively, effectiveness of parent-reported coping behaviors was partially linked to child health outcomes. Consistent with hypotheses, effectiveness of parent-reported coping behavior was significantly related to lung function. These results suggest a higher level of effectiveness of parent-reported coping behaviors are related to better lung function in children with CF. Additionally, although effectiveness of parent-reported coping behavior was not significantly associated with adherence, there was a
moderate, but meaningful effect size indicating higher scores of coping effectiveness may be associated with higher adherence.

The positive association between parental coping and child health outcomes found in the current study aligns well with previous research in pediatric cystic fibrosis indicating that parental ability to manage treatments are integral for the health outcomes of their children (Thompson et al. 1992; Eddy et al. 1998; Graetz et al. 2000; Anderson et al. 2001; Berge & Patterson 2004; DeLambo et al. 2004; Patterson, McCubbin, & Warwick, 1990; Quittner & DiGirolamo; Wong & Heriot, 2008). As a measure, the RISCS assesses both disease-specific coping behavior (e.g., response to medical procedures and symptoms, treatment adherence, eating/weight gain) as well as assessment of broader family function (e.g., response to sibling issues, parent finance issues, parent partner issues). This measure assesses both the broader parent role that requires general provision of emotional support, scheduling management and discipline in addition to disease-specific functioning. Therefore, the findings indicate that effectiveness of parent coping behaviors may be related to overall family functioning which supports findings that indicate that more effective family functioning protects against poorer lung function and other health outcomes (Everhart, Fiese, Smyth, Borschuk, & Anbar, 2014; Quittner, Opipari, Regoli, Jocobsen, & Eigen, 1992; Quittner, Espelage, Opipari, Carter, Eid, & Eigen, 1998). Given these findings, it will continue to be important to encourage the development of effective coping behaviors in parents of children with CF via supportive interventions promoting stress management and the development of coping skills (Quittner et al., 2016).
Association of Parent Mental Health, Coping, and Health-Related Quality of Life

No significant relationship was found between parent-reported psychological distress and HRQOL which differs from the limited number of studies assessing this association (Wong & Heriot, 2008; Brucefors, Hochwalder, Sjovall, & Helte, 2015). While it was hypothesized that higher parent-reported symptoms of depression and anxiety would predict lower child HRQOL, it is possible the null finding is an indicator that parent-reported ratings of child HRQOL may result in different scores from child-rated HRQOL. Specifically, prior concordance studies using the CFQ-R report higher parent-child concordance on physical domains (e.g., physical symptoms, digestive symptoms, respiratory symptoms) with lower concordance on non-observable domains (e.g., emotional functioning, body image, treatment burden)(Verrips et al., 2000; Eiser & Morse, 2001; Havermans, Vreys, Proesmans, & DeBoeck, 2006; Tluczek et al., 2013). It will be important for future studies to assess the relationship between parental psychological distress and child health outcomes, and include child-report data to continue to assess potential areas for intervention.

Alternatively, effectiveness of parent-reported coping behaviors was linked to the CFQ-R physical symptoms and vitality domains. However, counter to hypotheses, parents reporting more effective coping behaviors rated their children as having decreased HRQOL related to physical symptoms (e.g., walking, climbing, carrying objects) and vitality. Prior literature assessing coping via self-report questionnaires has found that parent coping effectiveness is related to child outcomes (e.g., mental health, adjustment, pulmonary functioning) with more effective coping being linked to better outcomes (Patterson, McCubbin, & Warwick, 1990; Wong & Heriot, 2008). The RISCs vignettes prompt parents to identify solutions to complete CF-related treatments. These
instructions could prompt parents to focus on treatment associations, including associations with poorer physical symptoms and challenges with treatment completion including oppositional behavior or avoidance. These associations may result in parents rating their child lower on HRQOL. Results from the current study may be indicative of parents identifying barriers to their child’s successful engagement (e.g., ability to participate in physical activities, ability to walk, ability to recover following physical exertion) which may prompt them to engage in more problem-focused coping behavior. It is also possible that parents utilizing more active and problem-focused coping may engage in higher levels of monitoring and increased involvement (e.g., overprotection) related to disease-specific as well as psychosocial situations their children face resulting in more negative psychosocial adjustment (e.g., Cappelli, McGrath, & MacDonald, 1989; Holmbeck et al., 2002). These high levels of engagement and monitoring may be associated with decreased feelings of autonomy and self-sufficiency in children which may result in lower vitality scores.

Despite the enormous treatment burden and shortened mortality, individuals with CF and their families have been shown to be tremendously resilient, with the majority of patients and parents reporting a high quality of life and normative levels of psychopathology (Szyndler et al., 2005). Additionally, Modi & Quittner (2003) found that children with a restricted range of disease severity limited the predictive validity of the CFQ-R. Therefore, results from HRQOL assessments may become more meaningful as the disease progresses or when assessing changes related to hospitalization or exacerbations.
Results from the current study indicate parents engage in more active problem-solving coping strategies when they are attending to poorer ratings of their children’s physical symptoms. While speculative, noticing differences in children’s respiratory function may have higher emotional valence for parents and prompt them to implement effective coping behaviors. Additionally, parents are reporting poorer affective ratings for their children. These parents may engage in increased monitoring to implement strategies ensuring ongoing treatment completion and thus be more likely to identify their children as more vulnerable. This supports prior research findings which indicate distressed parents of children with CF are more likely to report perceived child vulnerability (Hullman et al., 2010). It may be helpful to monitor degree of child engagement during clinic visits, elicit potential solutions to health-related challenges, and to assess child-specific coping behaviors to address potential differences in parent versus child perceptions. Discussing coping strategies and effective problem-solving may also provide the team with an opportunity to engage in strengths-based discussions with families, therefore continuing to promote resiliency.

**Associations of Depression and Anxiety Above versus Below Clinical Cut-offs**

While there were no statistically significant differences in child health outcomes between parents who reported symptoms above the mild or moderate clinical elevations for depression and anxiety, effect sizes indicate there may be potentially meaningful between group differences.

Effect size estimates indicate that there were meaningful differences in children’s rates of adherence based on parent-reported symptoms of depression. Specifically, parents who reported mild symptoms of depression had children with lower rates of adherence compared to parents who reported minimal depression symptoms, including
a small effect for differences in parents reporting mild symptoms compared to parents
not reporting clinically elevated symptoms. This finding fits well with previous research
indicating caregiver symptoms of depression are associated with lower adherence
(Modri, Driscoll, Montag-Leifling, & Acton, 2011; Quittner, Barker, Gellar, Butt, & Gondor,
2007; Smith et al., 2010; Hilliard et al., 2015). Conversely, a medium effect was found
associating higher levels of adherence with parents who reported symptoms of
depression in the moderate and above range. This may further support anecdotal
evidence suggesting parents prioritize adherence activities.

There were meaningful effect sizes for differences in lung function between
parents reporting symptoms of depression (medium effect) or anxiety (small effect)
above the mild cut-off. Counter to hypotheses, children of parents reporting clinically
elevated symptoms of depression or anxiety had higher lung function with the exception
of moderate anxiety that was associated with lower lung function. As previously noted,
due to exclusion criteria preventing families from participating during a CF exacerbation,
it is possible parents reporting symptoms of depression and anxiety may have recently
experienced a hospitalization or exacerbation resulting in improved lung functioning due
to intensive symptom management. Parents reporting high levels of anxiety may be
more vigilant with their child’s health that may prevent acute hospitalization for symptom
management; however, these children remain vulnerable to decreases in lung
functioning over time.

Parents reporting symptoms of depression above mild clinical elevations were
associated with small to moderate effect sizes on child HRQOL. These findings were
counter to hypothesis with results indicating that parents who reported elevated
symptoms of depression rated their children as experiencing higher HRQOL. While there are few studies evaluating the relationship between depression and HRQOL in children diagnosed with CF and their parents (Brucefors, Hochwalder, Sjovall, & Helte, 2015), other chronic illness populations have found links between elevated symptoms of depression and lower HRQOL (Janicke et al., 2007). Prior findings indicate fathers’ symptoms of depression may be more indicative of impact of child’s HRQOL (Brucefors, Hochwalder, Sjovall, & Helte, 2015). Therefore, it is possible this study did not find an association because mothers and fathers experiencing symptoms of depression may rate their child’s HRQOL differently. Specifically, mothers’ symptoms of anxiety and depression did not influence their ratings of child HRQOL while fathers experiencing elevated symptoms perceived their children to have lower HRQOL (Brucefors, Hochwalder, Sjovall, & Helte, 2015). While speculative, it is also possible parents experiencing higher levels of distress, both symptoms of depression and anxiety, may rate their children as experiencing improved HRQOL as a justification. Anecdotally, the RISCs responses indicate parents’ willingness to sacrifice self-care to ensure disease management and child functioning, despite personal costs (e.g., late to work).

Parents reporting symptoms of anxiety above mild clinical elevations were partially supported hypotheses and associated with small to moderate effect sizes for emotional functioning and vitality. Parents reporting symptoms in the mild clinical range rated their children as lower on HRQOL, while parents scoring in the moderate range rated their children higher on vitality. Parents experiencing symptoms of anxiety in the mild clinical range may engage in increased monitoring and negative child attributions. This may be beneficial for monitoring and managing disease-specific changes and
supports prior caregiver research in children diagnosed with asthma (Brown et al., 2006). Again, it is possible this study did not identify a significant relationship between parental psychological distress and child HRQOL due to gender differences. While speculative, these findings may indicate the importance of identifying differences between mild symptoms of distress from moderate to severe. These parents may be engaging in significantly different attributions, with parents experiencing higher levels of stress attempting to determine a justification for prioritizing their children’s HRQOL.

These findings indicate the importance of assessing parent-reported symptoms of psychological distress at clinic visits. This information may also be beneficial for providing tailored treatment interventions for parents reporting mild or moderate symptoms of distress.

**Limitations**

Although this study serves to address several gaps in the literature, it had several limitations. Due to the small sample size, there was limited power to detect meaningful relationships. Despite the small sample size, effect sizes indicate the importance of assessing the impact of parent-reported symptoms of depression, anxiety, and coping behaviors on child health outcomes. Self-report measures were used to assess symptoms of depression and anxiety that is associated with the potential risk for response bias. This study also relied on parent-report to assess child quality of life ratings. Concordance studies assessing parent and child agreement have been shown to be variable, particularly when parents attempt to rate relatively healthy children (Eiser & Moore, 2001). Previous findings show there is more agreement for observable functioning (e.g., physical symptoms) and less agreement for non-observable areas (e.g., emotional functioning) (Eiser & Moore, 2001). One study indicates there may be
caregiver differences in HRQOL ratings (e.g., fathers rate HRQOL more negatively) which was not assessed due to limited sample (Brucefors, Hochwalder, Sjovall, & Helte, 2015). Additionally, the children in this sample had mild illness severity at the time of measurement and therefore may not have experienced acute parenting stressors associated with having a child with increased disease severity. The homogeneity of the children’s health could have limited the ability to detect differences.

An important next step may be to assess the relationship between parent-reported psychological distress and lung function in a larger more representative sample of patients. There is also some evidence in the literature to support differences between fathers and mothers (Brucefors, Hochwalker, Sjovall, & Hjelte, 2015); however, due to sample limits this study could not assess potential differences related to caregiver gender.

**Clinical Implications and Future Directions**

Results from the current study provide evidence for the benefits of assessing psychological distress and coping behavior in parents of children diagnosed with CF. While a majority of the sample did not report clinically significant symptoms of psychological distress, a considerable proportion of parents indicate some elevation of symptoms of depression or anxiety. A recent consensus report includes recommendations for screening caregivers using PHQ-9 and GAD-7 measures and evaluating children for symptoms of anxiety and depression if their caregiver screens positively. Findings from the current study support incorporation of parent mental health screening as an ongoing part of clinic visits. These screenings would allow for subsequent intervention and more in-depth assessment of symptoms, as well as potential protective factors (e.g., effective coping, social support, etc.), as recommended
by the current consensus report. Additionally, this study provides support for the unique assessment of parent coping behavior. The effectiveness of parent coping is critical for successful disease management and may provide treatment team with opportunities to capitalize on family’s strengths as a way to increase treatment engagement. Additionally, it provides valuable information for intervention if the parents report engaging in highly ineffective coping behavior. The ability to facilitate improvements in the family system are likely to result in improvements in child health outcomes. Lastly, this study contributes to the CF literature by assessing the impact of parental psychological distress and coping on child HRQOL. This addressed a gap in the literature and provides valuable information for future studies to assess this relationship.

Future directions should continue to assess the patterns associated with parental symptoms of depression, anxiety, and effectiveness of coping behaviors. It will also be important for future studies to assess child mental health symptoms. This information will be helpful to continue studying the potential transactional effect of parent and child symptoms of psychological distress and ineffective coping strategies (Kazak et al., 2009; Quittner et al., 2014). It will be important for future studies to utilize longitudinal study designs to account for changes in parent mental health and coping behaviors over time. This may be helpful for capturing variations in parent-reported symptoms of psychological distress as they relate to declines or improvements in their child’s health status. Future longitudinal studies may also be able to determine associations between parent psychological distress and child health outcomes.
APPENDIX A
DEMOGRAPHIC QUESTIONNAIRE CYSTIC FIBROSIS AND FAMILIES BACKGROUND
INFORMATION QUESTIONNAIRE

Please complete the following pages based on the people who live in your home. We realize that all families are unique, and that caregivers may not necessarily be biologically related. If there is any confusion as to how best to answer the questions with regards to your family structure, please do not hesitate to ask and we will assist to the best of our abilities.

1. Zip code: ______________________
2. I am the child’s:  Mother   Father   Other____________________
3. Family structure:
   Mom’s age:_______

   Dad’s age:_______

4. Children in the home (including child enrolled in the study):

   Gender    Age    Grade in school
   __________________    ______    __________________
   __________________    ______    __________________
   __________________    ______    __________________
   __________________    ______    __________________
   __________________    ______    __________________
   __________________    ______    __________________
   __________________    ______    __________________

5. Others in the home (including extended family members):

   Gender    Age    Relationship to you
   __________________    ______    __________________
   __________________    ______    __________________
   __________________    ______    __________________
6. How many people living in the home smoke cigarettes? (please circle one)

None                  1 smoker in the home                  More than 1 smoker in the home

7. Where else is the child exposed to cigarette or tobacco smoke?

___________________________________________________________________________
___________________________________________________________________________
___________________________________________________________________________

8. Your marital status (please circle one):

1=Married                   4=Separated                   7= Single
2=Remarried after divorce   5= Remarried after widowed    8= Widowed
3= Divorced                 6= Cohabitating/living with partner

If separated, divorced, remarried, or widowed, please list child’s age at that time __________

9. Number of years married / cohabitating (living with partner) in current relationship _____

10. Number of people who are currently living in your household: _________

Please write your occupation:

11. Your occupation: ____________________________________________

12. Number of hours per week you work outside the home: ______________

13. Your partner’s occupation: ________________________________

14. Number of hours per week your partner works outside the home: ______________

15. Please circle your highest level of education completed.

1= less than 7th grade
2= junior high school
3= partial high school
4= high school graduate/GED
5= partial college or specialized training
6= college graduate
7= graduate or professional degree
16. Your racial background (please circle one):
1= African American 3= Hispanic 5= Native American
2= Caucasian 4= Asian 6= Other (please list) __________

17. Your child’s racial background (please circle one):
1= African American 3= Hispanic 5= Native American
2= Caucasian 4= Asian 6= Other (please list) __________

18. Please circle your partner’s highest level of education completed.
1= less than 7th grade 4= high school graduate/GED 6= college graduate
2= junior high school 5= partial college or specialized training
3= partial high school 7= graduate or professional degree

19. Your partner’s racial background (please circle one):
1= African American 3= Hispanic 5= Native American
2= Caucasian 4= Asian 6= Other (please list) __________

Please complete the following background information on your child with cystic fibrosis.

20. How many times has your child been hospitalized for cystic fibrosis in the past year: ______

21. How many times has your child been hospitalized for non-cystic fibrosis related issues over the past year: ______

22. How many days of school has your child missed for cystic fibrosis-related problems over the past year: ______

23. How many days of work have you missed due to your child’s cystic fibrosis over the past year: ______

24. How many days of work has your partner missed due to your child’s cystic fibrosis over the past year: _______
25. List all medications your child is currently taking:

________________________________________________________________________

________________________________________________________________________

________________________________________________________________________

________________________________________________________________________

________________________________________________________________________

________________________________________________________________________
Chart Review:

Age: ____________

Mutation type: ____________

**BMI**

Height: ____________

Weight: ____________

BMI: ____________

BMI percentile: ____________

**PFT**

FEV$_1$: ____________

Percent predicted: ____________

FVC: ____________

Percent predicted: ____________

FEF$_{25-75}$: ____________

Percent predicted: ____________

TLC: ____________

Percent predicted: ____________
Current medications:

<table>
<thead>
<tr>
<th>Medication</th>
<th>Dose prescribed</th>
<th>Frequency (i.e. 2x per day)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
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<tr>
<td></td>
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<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Dates of Pulmonary Clinic visits in the past year:

______

______

______

Dates of ED visits in the past year:

______

______

______

Dates of CF-related hospitalizations in the past year:

______

______

______

Dates of CF-related operations in the past year:

______

______

______

Dates of Courses of antibiotics in the past year:

______________

______________
Comorbid Diagnoses/Problem List:

______________________________________________________________________

_______

______________________________________________________________________

_______

Previous sleep study? Yes / No

If yes, dates:

______________________________________________________________________

_______

______________________________________________________________________

_______
Appendix B

QUESTIONNAIRES 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)

<table>
<thead>
<tr>
<th>Problem</th>
<th>Not at all</th>
<th>Several days</th>
<th>More than half the days</th>
<th>Nearly every day</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Little interest or pleasure in doing things</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>2. Feeling down, depressed, or hopeless</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>3. Trouble falling or staying asleep, or sleeping too much</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>4. Feeling tired or having little energy</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>5. Poor appetite or overeating</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>6. Feeling bad about yourself — or that you are a failure or have let yourself or your family down</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>7. Trouble concentrating on things, such as reading the newspaper or watching television</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>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</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>9. Thoughts that you would be better off dead or of hurting yourself in some way</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
</tbody>
</table>

For office coding

\[ 0 + \square + \square + \square \]

= 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?

<table>
<thead>
<tr>
<th>Difficulty Level</th>
<th>Not difficult at all</th>
<th>Somewhat difficult</th>
<th>Very difficult</th>
<th>Extremely difficult</th>
</tr>
</thead>
</table>
Generalized Anxiety Disorder- 7-item (GAD-7):

<table>
<thead>
<tr>
<th>Over the last 2 weeks, how often have you been bothered by the following problems?</th>
<th>Not at all sure</th>
<th>Several days</th>
<th>Over half the days</th>
<th>Nearly every day</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Feeling nervous, anxious, or on edge</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>2. Not being able to stop or control worrying</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>3. Worrying too much about different things</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>4. Trouble relaxing</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>5. Being so restless that it's hard to sit still</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>6. Becoming easily annoyed or irritable</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>7. Feeling afraid as if something awful might happen</td>
<td>0</td>
<td>1</td>
<td>2</td>
<td>3</td>
</tr>
</tbody>
</table>

Add the score for each column

Total score (add your column scores) =

If you checked off any problems, how difficult have these 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
RISCs Vignettes

Vignette 1
You are all sitting down at the table for dinner, and your children start arguing and teasing each other. You ask them to stop, but they start fighting again in a few minutes. You would like to have some peace and quiet during dinner. What would you do or say in this situation?

Vignette 2
You are in the middle of doing treatment with your son/daughter. Your other child starts jumping around and wants you to play a game now and says, “you never do anything with me. You like him/her better than me.” You try to be as fair as you can, but doing treatment is important and takes a lot of your time. What would you do or say in this situation?

Vignette 3
Your son/daughter is in the hospital. Your other child is upset about it and says, “I really miss him. I’m scared he’s not coming back.” You want to be honest, but you’re not sure how much to say to your child about CF. How would you handle this situation?

Vignette 4
You are at the clinic for your child’s check-up, and you’ve been sitting in different waiting rooms since early this morning. Now you are in the X-ray Department and have already waited for over an hour. You are tired and your child is restless and bored, but they still haven’t called your name. How would you handle this situation?

Vignette 5
Every year you sit down with the new teacher about CF, and how it affects your child. Today, your son was upset because the teacher wouldn’t let him go to the bathroom, and asked him to wait until recess. You’re frustrated because you’ve already discussed these issues and wish the teacher could be more sensitive. How would you handle this situation?

Vignette 6
You have to run some errands and need your child to come with you. About ten minutes ago, you told him/her to get ready and turn off the T.V. You want to leave now and he/she is still lying on the floor watching T.V. You say, “Hey, let’s get going!” He/She keeps stalling and then starts whining. Lately, he/she never seems to listen to you, and you’re getting frustrated. What would you say or do in this situation?

Vignette 7
You’ve brought your child into clinic because he/she has been coughing a lot at night and seems tired. Now, the tests show his/her pulmonary functions are down. You want a clear treatment plan, but the doctor is vague about what you should do. It seems like he’s not listening to your questions, but is just responding in a routine way. What would you say or do in this situation?
Vignette 8
Your child asks for a snack in the evening. You say, “Have some cheese and crackers, but be sure to take your enzymes!” Later, you go into the kitchen and he/she has finished the snack but has forgotten to take enzymes. Your child says, “It’s just a snack don’t really need them.” You are tired of constantly reminding your child and wish he/she would be more responsible. What would you say or do in this situation?

Vignette 9
You are waiting to be reimbursed for medications you paid for several months ago. The insurance company always seems bogged down in red tape, and you are not sure how to speed things up. You have a lot of expenses to cover this month and could really use the money. How would you handle this situation?

Vignette 10
The nurse is trying to start an IV on your son/daughter and you know it might take a few tries. When the nurse is getting ready, you son/daughter starts crying and tells you he/she is scared. You feel helpless and it hurts you to see him/her going through these painful procedures. What would you say or do in this situation?

Vignette 11
You know it’s important for your son/daughter to eat healthy meals, so you’ve fixed a dinner you think he/she will really like. Your son/daughter sits down at the table, takes one bit and say, “I don’t like this. I don’t want to eat it.” It seems like it’s impossible to please him/her. What would you say or do in this situation?

Vignette 12
It’s another hectic morning. Everyone has to be out the door and on their way by 8:00 am. You’re behind schedule and are worried that if you do the mist and percussion plus everything else you need to do, you will be late for work and your child will be late for school. You’re frustrated because there is always too much to do in the morning. What would you say or do in this situation?

Vignette 13
You and your son/daughter are shopping at the grocery store. He/She starts to cough and you notice people are staring. Someone says, “that child sure sounds sick. Should he/she be at home in bed?” You’re embarrassed and tired of having to explain his/her coughing to stranger. What would you say or do in this situation?

Vignette 14
You and your partner are planning to go to a special dinner next week. You usually have a relative babysit, but everyone is busy that particular night. You really want to go, but you’re worried about finding someone who is responsible enough to handle your child’s medications and treatment. How would you handle this situation?

Vignette 15
Your child has ball practice today. On the way there, he/she asks to go over to a friend’s house after practice. It’s always hard getting treatment and everything else done in the evening.; You want him/her to be able to do everything other kids do, but getting treatment done is important for his/her health. What would you say or do in this situation?

Vignette 16
You let your son/daughter stay up later than usual last night to watch a movie. This morning you have already told him twice to get up. How it’s so late you’re worried that he/she might miss the school bus. You say again, “Get up!” but he/she still doesn’t budge. What would you say or do in this situation?

Vignette 17
Recently your child has been coming home from school with stomach aches. The school nurse is supposed to give him/her the enzymes before lunch, but you find out they’re being given at the wrong time. You’re upset because you carefully explained to the nurse why the enzymes must be given on schedule. It’s hard to trust other people with something this important. How would you handle this situation?

Vignette 18
Your child has been in the hospital for over a week. You want to be there as much as possible, but you also need to keep things going at home. Traveling back and forth to the hospital, and trying to fit in work, chores, and time with your family is exhausting. Everyone in the family feels stressed. How would you handle this situation?

Vignette 19
The doctor has just prescribed several new medicines for your child. You know your insurance will only cover part of it and it’s going to cost you several hundred dollars out of your own pocket. You already have lots of bills to pay this month. You want your child to have the best possible care, but you’re not sure how you can pay for all of it. What would you say or do in this situation?

Vignette 20
Before breakfast you give your child his/her enzymes like you do every morning. He/she surprises you by asking, “why do I have these? No one else does. Will I always have to take them?” What would you say or do in this situation?

Vignette 21
You’re reading the newspaper when you come across an article about a child with CF who recently died. The child was just a few years older than your own. Your son is doing really well right now, but you’re reminded that it may not always be like this. You try not to think about it but you can’t seem to get it off your mind. How would you handle this situation?

Vignette 22
Your child comes home from school and asks if he/she can go outside to play. You know he has homework to do, and he/she is always trying to get out of it. You would like to give him a break from school, but you know unless the homework is done now, it will be hard to fit it in later. What would you say or do in this situation?

Vignette 23
Your child is in the hospital for a tune-up. When you visit, his/her dinner is sitting on the tray untouched. He/she was afraid to eat because nobody brought any enzymes, even though he/she asked for some. Last night, you watched one of the staff do the percussion and didn’t think it was very thorough. You are upset that he/she’s in the hospital, but not getting proper care. What would you say or do in this situation?

Vignette 24
Your son/daughter came home from school excited because a friend invited him/her to spend the night. He/she hasn’t spent the night away from you before, and you don’t know the parents very well. You’re worried about how he/she will get the enzymes and treatment he needs. He/she really wants to go, but you’re just not sure. What would you say or do in this situation?

Vignette 25
You tell your child, “It’s time to start treatment!” He/she refused to come over and starts to fuss. He/she says, “NO! I don’t want to do it. I’m not going to do it.” He/she always wants to get out of doing treatment and you find it frustrating. What would you say or do in this situation?

Vignette 26
Your son/daughter comes home from school and seems upset. When you ask what’s wrong, he/she tells you that the other kids are teasing him/her about coughing a lot and are saying that they can catch his/her disease. You can see how hurt your child is. What would you say or do in this situation?

Vignette 27
The doctors have said that your son/daughter needs to gain weight. You’ve been trying hard to get him/her to eat more. The next clinic visit is coming up soon and he/she isn’t gaining any weight. You’re worried about what the doctors will say and you wonder if there’s anything else you can do. How would you handle this situation?

Vignette 28
Several times this week you have told your son/daughter to clean his/her room. It’s really a mess and you’re tired of always nagging him/her about doing chores. You remind him/her again and he/she says, “I’ll do it later.” What would you say or do in this situation?

Vignette 29
You and your partner have both had a busy day and are exhausted. It’s time to do your child’s mist and percussion treatment, but you disagree about who should do it.
This issue comes up a lot, but never seems to get resolved. Something you feel like your partner isn’t doing as much as you are. What would you say or do in this situation?

Vignette 30
A few weeks ago, you and your partner agreed to cut back on expenses. Now, your partner comes home with something you really don’t need and can’t afford. You thought you had agreed not to spend extra money like this. What would you say or do in this situation?

Vignette 31
There is a class you really want to take that meets two evening a week. You talk about it, but your partner is not very supportive, and doesn’t want to be responsible for the kids on those two nights. You would like to be able to go. What would you say or do in this situation?
Cystic Fibrosis Questionnaire - Revised (CFQ-R):

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

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 your 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


Page 1
### CFQ - R

**Cystic Fibrosis Questionnaire - Revised**

**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.

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

Please check the box matching your response.

**During the past two weeks, indicate how often your child**

<table>
<thead>
<tr>
<th></th>
<th>Always</th>
<th>Often</th>
<th>Sometimes</th>
<th>Never</th>
</tr>
</thead>
<tbody>
<tr>
<td>6. Seemed happy</td>
<td></td>
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<tr>
<td>7. Seemed worried</td>
<td></td>
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<tr>
<td>8. Seemed tired</td>
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<tr>
<td>9. Seemed short-tempered</td>
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<td>10. Seemed well</td>
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<tr>
<td>11. Seemed grouchy</td>
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<tr>
<td>12. Seemed energetic</td>
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<tr>
<td>13. Was absent or late for school or other activities because of his/her illness or treatments</td>
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</tbody>
</table>

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:

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

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.

<table>
<thead>
<tr>
<th>Question</th>
<th>A great deal</th>
<th>Somewhat</th>
<th>A little</th>
<th>Not at all</th>
</tr>
</thead>
<tbody>
<tr>
<td>33. My child had trouble gaining weight</td>
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<tr>
<td>34. My child was congested</td>
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<td>35. My child coughed during the day</td>
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<td>36. My child had to cough up mucus</td>
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<tr>
<td>37. My child’s mucus has been mostly:</td>
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<tr>
<td>□ Clear</td>
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<tr>
<td>□ Clear to yellow</td>
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<tr>
<td>□ Yellowish-green</td>
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<tr>
<td>□ Green with traces of blood</td>
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<tr>
<td>□ Don’t know</td>
<td></td>
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</tbody>
</table>

During the past two weeks:

<table>
<thead>
<tr>
<th>Question</th>
<th>Always</th>
<th>Often</th>
<th>Sometimes</th>
<th>Never</th>
</tr>
</thead>
<tbody>
<tr>
<td>38. My child wheezed</td>
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<tr>
<td>39. My child had trouble breathing</td>
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<td>40. My child woke up during the night because he/she was coughing</td>
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<td>41. My child had gas</td>
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<tr>
<td>42. My child had diarrhea</td>
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<tr>
<td>43. My child had abdominal pain</td>
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<tr>
<td>44. My child has had eating problems</td>
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</tbody>
</table>

Please be sure you have answered all the questions.

THANK YOU FOR YOUR COOPERATION!
REFERENCES


BIOGRAPHICAL SKETCH

Alyssa Fritz earned her B.A. in psychology—with a double major in history—from the University of North Carolina Wilmington in 2009. She subsequently continued at UNC Wilmington, earning an M.A. in psychology. Her master's thesis research was on the topic of child witnesses of intimate partner violence. In 2012 she began studying clinical health psychology at the University of Florida as part of Dr. Stephen Boggs’s Child Study Lab and later as part of Dr. David Fedele’s Pediatric Behavioral Health Lab. Her research interests are focused on the area of family functioning in chronic illness, particularly the interaction of parent mental health and child treatment adherence.