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Illness Identity, Experience, and Perceptions as Longitudinal Predictors of Adherence Behaviors and Health Outcomes in Adolescent Cystic Fibrosis

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Illness Identity, Experience, and Perceptions as Longitudinal Predictors of Adherence Behaviors and Health Outcomes in Adolescent Cystic Fibrosis

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Dedication

For Emily.

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Abstract

Illness Identity, Experience, and Perceptions as Longitudinal Predictors of Adherence Behaviors and Health Outcomes in Adolescent Cystic Fibrosis

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People with Cystic Fibrosis (CF) experience considerable symptom and treatment burden on a daily basis. Airway Clearance Therapy (ACT) is one of the most time-consuming treatments and has low rates of adherence. Adherence behaviors are important to address in adolescence, as patients begin to take over responsibility from parents. However, adolescents with CF experience unique barriers to treatment adherence. Their symptoms and treatments can disrupt developmentally-appropriate pursuits like peer relationships and affect identity development. *Illness identity* describes distinct ways in which chronic illness can be integrated into a patient's identity, with varying effects on mental health and treatment adherence. Patients' experience of CF also alters their treatment adherence by affecting beliefs about the necessity and efficacy of their treatments. As such, improving treatment adherence may require considering not only the adolescents' illness- and treatment-related beliefs, but also the experiences that have informed those beliefs. This study of 120 adolescents with CF recruited from a Central

Texas medical center uses regression to determine whether past illness-related health, represented by average pulmonary function, predicts ACT adherence, and whether illness identity and treatment beliefs (*Beliefs*) mediate the relationship. The study also uses regression to assess whether ACT adherence mediates the relationship between past *Beliefs* and future pulmonary function. This study would allow clinicians to identify adolescents with uncontrolled illness-related health as at higher risk for developing future maladaptive *Beliefs* that could affect treatment adherence. It would also provide guidance on targeted interventions for adolescents with chronically poor treatment adherence that take their unique developmental stage into account.

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INTRODUCTION

Few of the myriad experiences people have in their lifetime are understood in objective terms. Instead, two people experiencing the same event might have completely distinct interpretations of why the event occurred and what it means. This subjective appraisal of one's experience is informed by the beliefs that have been generated and reinforced, or disconfirmed, by past experiences. Indeed, Erik Erikson posited that our very identities are formed through the interaction of our subjective appraisals of our own actions and our experiences (1950, 1968). However, our subjective appraisals of objective information influence more than our self-esteem; they influence, and can change, our very understanding of reality. For example, people with chronic illness have varying beliefs about the severity of their illness and the efficacy of their prescribed medications (Bucks et al., 2009; Horne et al., 2013; Sherman et al., 2019). Those perceptions influence their treatment adherence behaviors and their resulting physical health outcomes (Hilliard et al., 2015). This is an especially pressing concern for adolescents who are often just beginning to take over responsibility for their treatments from parents. Appraisals of oneself and the world play an especially pressing role in adolescence, when rapid physical and cognitive advancements incentivize adolescents to develop their identities (Erikson, 1968).

Adolescents with chronic illness often have an additional dimension of identity development to contend with: how their illness will fit into their still-developing identities. The manner in which adolescents integrate their illness into their identities has important implications for how they manage their disease and their mental health outcomes (Oris et al., 2016). Disease management is an especially salient concern for chronic illnesses that require aversive, numerous, and lengthy treatment regimes. Cystic Fibrosis (CF) is one such illness in which quality of life, and the likelihood of survival, can be changed

dramatically by completing treatments (Eakin et al., 2011). However, many adolescents with CF have difficulty adhering to their treatment regimes (Smith et al., 2010; White et al., 2009). Their adherence is further complicated by the fact that they are often forced to choose between spending time with peers and engaging in treatments that further accentuate how they differ from peers (Askew et al., 2017; Ernst et al., 2010; Sawicki et al., 2014). As such, many adolescents with CF may have complex feelings about how their illness fits into their identity.

The Illness Identity Questionnaire (IIQ; Oris et al., 2016) is a novel measure that describes four manners of identification with one's illness: Acceptance, Rejection, Engulfment, and Enrichment. These manners of illness identification are intertwined with beliefs about one's illness and treatments and can affect treatment adherence behaviors. These beliefs about one's illness and treatments are important intervention targets (Nguyen et al., 2016; Petrie et al., 2012), and the instability in perceptions of the self and world in adolescence create an opportunity to intervene and change unhelpful beliefs, leading to better health outcomes.

INTEGRATIVE ANALYSIS

Chronic Illness in Adolescence

Chronic illness can interfere with normal developmental processes such as hormonal development and growth, sexual maturation, appearance, school attendance, access to peers, social development, life prospects, and conceptions of self. Each way in which chronic illness makes an adolescent's life deviate from that of their peers creates additional psychosocial issues that may complicate their willingness to accept their illness as a part of their lives. Research on medication adherence in chronically ill adolescents and youth has shown that not all interventions with known efficacy for adults work with adolescents, lending partially to the unique developmental challenges posed by their developmental stage in life (Dean et al., 2010; Suris et al., 2004).

Another factor affecting adolescent medication adherence is related to a practical expression of identity development in adolescence—increasing independence. Many chronically ill adolescents have to rely on their families due to physical, logistical, or financial limitations imposed by their illness or their medication regimes. This complicates their ability to establish the independence typical to adolescence. Chronically ill adolescents often begin to establish some degree of independence by taking over control of their medications, often with poor results (García-Pérez et al., 2013; Keough et al., 2011; Kyngäs et al., 2000; Kyngäs & Rissanen, 2001). Simple education about their illness and the importance of adhering to medications alone does not appear to be a robust catalyst of adherence change for adolescents, or adults (Dean et al., 2010; Norris et al., 2001). Instead, changing attitudes and motivations towards treatments and illnesses in general may be a more meaningful way to increase adherence to medications. This is an especially important concern for those with lengthy, aversive treatment regimes, such Cystic Fibrosis patients.

Introduction to Cystic Fibrosis

According to the Cystic Fibrosis Foundation (CFF) Patient Registry, there are an estimated 30,000 people currently diagnosed with Cystic Fibrosis (CF) in the United States (Cystic Fibrosis Foundation, 2017). People with CF experience dysfunction in multiple organ systems that, despite the best available treatments, ultimately lead to an early death with a life expectancy of under 45 years as of 2018 (Cystic Fibrosis Foundation, 2019). People with CF inherit a mutated Cystic Fibrosis Transmembrane Conductance Regulator (CFTR) gene from each parent that affects their sweat glands, lungs, pancreas, intestines, liver, and reproductive organs (Giddings, 2009; Teichroew, 2016). The CFTR mutation impedes the movement of chloride within these organs' cells which prevents them from attracting water to their mucus-covered exteriors. Without sufficient water content the mucus becomes overly sticky and viscous and hinders proper organ function. Various CFTR allele mutations exist, and people can inherit different combinations of mutations from their carrier parents. Different CFTR mutations have various degrees of impairment in chloride movement, so the severity of CF impairment and prognosis can often be determined by the specific mutations inherited.

When the CFTR gene is sufficiently impaired, it causes widespread dysfunction within the body. In the lungs, excessive mucus can block airways, impair breathing, trap harmful bacteria, and lead to infection, inflammation, and eventually respiratory failure. Although a small number of individuals are able to receive lung transplants, respiratory failure causes nearly 63% of deaths in CF (Cystic Fibrosis Foundation, 2018; Giddings, 2009). Roughly 30% of people with CF also experience asthma as a result of chronic lung inflammation (Cystic Fibrosis Foundation, 2019). Lung function is considered an important marker of CF severity and is often considered to reflect health outcomes, the

efficacy of medications, and the degree of treatment adherence (Szczesniak et al., 2017). Lung function is measured by forced expiratory volume in 1 second (FEV1), the percent of air expelled in one second relative to a healthy peer, where higher percentages indicate greater health.

In the pancreas of people with CF, thickened mucus affects the secretion of digestive enzymes that the intestines need to absorb nutrients from food. The intestines' inability to absorb nutrients can affect growth and the ability to gain weight, often resulting in smaller stature (Teichroew, 2016). Body Mass Index (BMI) and weight are also indications of severity. Increasing both are important and common treatment goals, as maintaining a healthy weight is vital to pulmonary function. Prolonged pancreatic dysfunction in CF patients can also lead to insulin-dependent Cystic Fibrosis-Related Diabetes (CFRD) in roughly 30% of adults (Cystic Fibrosis Foundation, 2018). Furthermore, low water content in the liver causes abnormal bile which can affect digestion, fat absorption, and the filtration of harmful toxins from the blood (Giddings, 2009; Kobelska-Dubiel et al., 2014). The thickened bile clogs bile ducts and leads to chronic liver inflammation, which can lead to cirrhosis and even death. In addition to pulmonary and digestive disruption, CF often results in male infertility due to the structural abnormality or absence of the vas deferens, though surgical procedures can often restore function (Giddings, 2009). While most women with CF can become pregnant, pregnancy can worsen the symptoms of CF. Other complications with CF can include frequent infections, fatigue, low blood pressure, elevated heart rate, gallstones, irregular bowel movements, osteoporosis, and urinary or fecal incontinence related to weakening of the pelvic floor from excessive coughing (Giddings, 2009; Neemuchwala et al., 2017; Teichroew, 2016).

This complex range of symptoms can cause frequent, widespread disruptions to health and activities of daily living. Fortunately, modern treatments for CF can significantly lessen symptom burden and prolong lifespans. However, such treatments are complex, time-consuming, and often aversive or burdensome. The needs imposed by complex treatments, as well as by CF itself, can also interfere with normal developmental processes during adolescence such as seeking independence from parents and establishing peer relationships (Ernst et al., 2010; Suris et al., 2004). Some treatments can even hinder physical and sexual maturation during puberty. These and other barriers contribute to poor treatment adherence during adolescence which often translates to poorer adult adherence (Bucks et al., 2009; Sherman et al., 2019). Adolescence is an important time to establish many vital health behaviors, and is therefore an important intervention target to prevent poor adherence in adulthood. However, addressing the problem of treatment adherence requires careful consideration of the historical context, psychosocial sequelae, treatment burden, and internal experience of having CF, as well as how those factors intersect with the developmental stage of adolescence.

HISTORICAL CONTEXT OF CYSTIC FIBROSIS PROGNOSIS AND TREATMENT

While CF was historically diagnosed in childhood, newborn screening has gradually become the norm in obstetric and pediatric settings and has resulted in improved health outcomes through earlier detection and treatment (Bender et al., 2011; Cystic Fibrosis Foundation, 2017; Grosse et al., 2005; Ross, 2008). Newborn screening in the United States (US) began in 1982 with a single clinic in Colorado, and expanded exponentially after the Centers for Disease Control (CDC) and the CFF provided a joint recommendation for newborn testing in each state in 2003 (Grosse et al., 2005; Ross, 2008). By 2010, all 50 states in the US were using newborn screens for CF (Bender et al.,

2011). According to the CFF, 62.4% of the nearly 900 new CF patients in 2016 were diagnosed through newborn screening, compared to only 9.2% in 2001. Increased screening, earlier detection, and improvements in treatments have increased the predicted median lifespan for individuals with cystic fibrosis from roughly 33.3 to 44.4 years old from 2000 to 2018 (Cystic Fibrosis Foundation, 2016, 2019). In 2018, 60% of patients were eligible for a medication that addresses the root cause of CF and dramatically improves health outcomes and prognosis (Cystic Fibrosis Foundation, 2019). By 2019, that number had risen to 90% (Middleton et al., 2019). Although the number of children and adolescents with CF has remained somewhat stable from 1986 to 2016, the percentage of the CF population made up by adults has increased from under 30% to over 54% during that time. This is because over half of individuals with CF are now living well into adulthood. The number of adults with CF who reported being married or in a serious relationship has nearly tripled from 1998 to 2018, while the number of people with Bachelor's or advanced degrees has nearly quadrupled (Cystic Fibrosis Foundation, 2019). A prolonged lifespan now allows them to participate in activities of adulthood that may have been less attainable before breakthroughs in treatments improved their life-span and quality of life. However, given that many children and adolescents with CF may not have expected to live past early adulthood prior to the relatively recent advances in treatment, this emerging adult CF population faces unique health and psychosocial challenges. One such challenge is treatment adherence.

CURRENT TREATMENTS FOR CYSTIC FIBROSIS

Many children and adolescents have difficulty maintaining adherence to CF's complex treatment regime, and these behaviors have often been shown to continue into adulthood if left unaddressed (Bucks et al., 2009; Sherman et al., 2019). While parents

typically take responsibility for their younger children's treatment adherence, youth tend to start taking over control of their treatments by adolescence, sometimes with poor results. Consistent adherence to treatments is often the only way for people with CF to maintain bodily health and prolong their lives (Eakin et al., 2011; Eakin & Riekert, 2013). Therefore, adolescence, when caretakers still have some degree of influence and ability to support patients, is a vital time to establish and improve adherence behaviors.

Part of the difficulty in increasing treatment adherence in adolescents, and hopefully in the adults they will grow into, is due to the complexity of treatments themselves. Studies of treatment burden have found that people with CF are prescribed a median of 7 daily treatments that amount to an average of at least 108 minutes per day, though many spend even more time (Sawicki et al., 2009). Treatments for CF include oral medications for the gastrointestinal system, inhaled nebulized medications for the pulmonary system, physical methods to extract excessive mucus from the lungs, and gene therapies that directly address the CFTR mutation's dysfunction. This review will focus on a subset of pulmonary therapies, Airway Clearance Techniques (ACT).

Airway Clearance Therapies dislodge mucus from airways to allow for easier breathing. Chest physiotherapy, a popular form of ACT, involves the rapid external movement of the chest cavity to dislodge mucus in the lungs, which is then expelled by coughing. Nearly 80% of people with CF use a mechanical high-frequency chest wall oscillation (HFCWO) vest to aid in dislodging the mucus. The CFF also recommends the use of exercise as an additional method of ACT which roughly 40 to 50 percent of adolescents and young adults report using (Cystic Fibrosis Foundation, 2019). The efficacy of ACT is also improved through the use of the aforementioned inhaled medications that thin mucus in the lungs and dilate the airways. People with CF may be prescribed ACT 2

or more times per day for a duration of at least 20-40 minutes each time (Sawicki et al., 2009).

Treatment Adherence

Each prescribed treatment requires different degrees of preparation, time, energy, equipment, and privacy. Some treatments are considered more aversive and burdensome, or create significant inconvenience to users' sense of normalcy or social belonging. The latter is an especially important concern for adolescents (Sawicki et al., 2009, 2014). As a result, different treatments have different rates of adherence, particularly as adolescents and young adults begin to take control of their own care (Eakin et al., 2011). However, given that treatments only work properly when sufficiently adhered to, understanding and improving the barriers to adherence is an important goal for mental and physical health providers. The first step to understanding barriers to adherence is to understand how adherence is assessed.

MEASUREMENT

Treatment adherence must be accurately measured before it can be understood or improved. There are various challenges to accurately collecting data on treatment adherence for kids and adolescents with chronic illnesses in general, and for CF specifically. Countless studies have documented significant differences between parent and child reports about treatment adherence for various illnesses. Within CF, Goodfellow and colleagues (2015) found that children with CF were significantly more likely than their parents to report low adherence to enzyme supplements. The lower report may be more accurate as enzyme supplements are a treatment that children would likely be responsible for self-administering during mealtimes at school, which precludes parental supervision.

Parents have also reported higher adherence than kids and adolescents for other treatments (T. White et al., 2009). In addition to discrepancies between child and parent reports of adherence, there are often differences between subjective reports of adherence, such as patient interviews, medication diaries, or validated self-report questionnaires, and objective measures, such as Medication Event Monitoring Systems (MEMS), pill counts, or pharmacy refill data.

Patient interviews may differ even further from objective measures than other subjective measures like validated self-report adherence measures or medication diaries (Garber et al., 2016). Even so, a systematic review of studies comparing subjective and objective adherence measures across chronic illnesses found that only 23.3% of studies reported significant correlations between validated self-report questionnaires and MEMS data (Monnette et al., 2018). Another study measuring CF patients' adherence to CFTR modulators found no significant correlations between self-reported adherence, electronic monitoring measures, or pharmacy refill data (Siracusa et al., 2015). Similarly, while ACT often involves a large, noisy mechanical vest which should make it easier for parents to determine whether it is in use, one study found that parent-reported adherence to ACT identified 66% of the sample as high adherers, while MEMS data identified only 35% (Oates et al., 2019). Another study found that parents were more likely than clinicians to rate their children's ACT adherence as high (51% vs 35%; Eddy et al., 1998).

Similarly, self-reported, parent-reported, and clinician-reported adherence to inhaled medications have been demonstrated to be biased towards overestimation compared to MEMS data (Daniels et al., 2011). As such, while there is no gold standard for measuring treatment adherence in CF patients, partially due to the variety and quantity of treatments, more objective measures are preferable. At minimum, the more consistent

use of objective measures would enable easier comparisons between studies attempting to improve adherence through various means.

Prevalence of Poor Adherence

Even if each treatment for CF could be measured using an objective, accurate method, the adherence rate for different treatments would still differ. Some treatments are adhered to less due to the time they take, their aversiveness, or the logistical challenges of completing them. Overall, treatment adherence rates appear to vary from roughly 30 to 70 percent across all treatments (see *Table 1*). However, considering that ACT is one of the more aversive and vital components of the treatment regime, this review will focus exclusively on adherence rates for ACT.

Of the myriad treatments for CF, Airway Clearance Therapies (ACT) are often considered to be the most cumbersome, aversive, and time-consuming. Having to perform at least 30 minutes of ACT per day is associated with the most severe experience of treatment burden (Sawicki et al., 2009). In one study, 41% of people with pediatric CF reported completing less than half of their prescribed airway clearance therapies (Smith et al., 2010). This included the 26% who reported completing none of their treatment. On average, participants reported completing 54% of prescribed ACT's and used them for 68% of the prescribed duration (Smith et al., 2010). Another study of young adults with CF found that only 29.5% reported that ACT was a part of their daily routine and 11.4% never adhered to ACT (Myers & Horn, 2006). A study using an objective measure of pediatric adherence to ACT, high-frequency chest wall oscillation (HFCWO) vests that automatically recorded use, found that only 34.6% of the sample adhered 80% of the time or more. While the average adherence for the sample was 60%, over a quarter of the sample (28.2%) completed less than 35% of their treatment (Oates et al., 2019).

Factors Affecting Adherence In Chronic Illness

One issue that may affect both measured and actual adherence rates is the fact that prescribed treatments for CF tend to be complex and lengthy, and can vary in intensity both between patients and between treatments. As such, adherence rates often vary according to both treatment and patient characteristics. In particular, pediatric adherence tends to worsen as age increases, parental influence decreases, and as treatment complexity and burden increase. Finally, the beliefs people have about their illnesses, their treatments, and themselves also affect their treatment adherence.

DEMOGRAPHIC

Various demographic factors affect treatment adherence in CF populations. Generally, adherence tends to worsen with greater age, lower socio-economic status (SES; Smith et al., 2010), and minority status (McGarry et al., 2017).

Age

In pediatric CF, and other chronic illnesses, older age has been repeatedly shown to be associated with poorer treatment adherence. In adolescents with diabetes, older age has been associated with poorer treatment adherence and health outcomes (Oris et al., 2016). Age has also predicted weaker beliefs in treatment efficacy, which in turn predicted worse adherence to dietary self-care behaviors (Nouwen et al., 2009). Within pediatric CF, older children have significantly poorer self-reported adherence to various treatments than younger children (Everhart et al., 2014; Smith et al., 2010; Zindani et al., 2006). Specifically, older age has been associated with poorer adherence to ACT, antibiotics, and vitamins (Bucks et al., 2009; Goodfellow et al., 2015). Older age is associated with weaker beliefs about whether ACT is a necessary part of their treatment (Bucks et al., 2009).

Similarly, a study that measured adherence to vitamins through electronic pill bottle caps that automatically recorded when medication was accessed found that adolescents with CF took a significantly lower proportion of prescribed multivitamins than kids (Zindani et al., 2006). The worsening of pediatric adherence over time appears to continue into adulthood. A study using electronic monitoring of nebulized medication use by adults with CF found a median adherence rate of just 36% where the middle 50% of participants' adherence ranged from 5% to 84.5% (Daniels et al., 2011).

Socioeconomic Status

Socioeconomic status (SES) is often described in terms of factors like income, wealth, and the financial ability to pursue higher education. In general, lower SES has been associated with lower health-related quality of life in people with CF (Quittner et al., 2010). Lower parental education has also been associated with less adherent children (Smith et al., 2010). Low SES may impact treatment adherence by affecting familial stress, time, transportation, healthcare access, and in some cases information about treatments.

Race or Ethnicity

While the majority of individuals in the CFF's 2018 Patient Registry identified as Caucasian, 4.7% and 9.4% identified as African American and Hispanic respectively (Cystic Fibrosis Foundation, 2019). Of note, the proportion of Hispanic-identifing people with CF nearly doubled from 2003 to 2018 (Cystic Fibrosis Foundation, 2019). While the percentage of minorities with CF is comparatively smaller, CF's effects on these communities should not be discounted, especially considering that they often suffer worse health and psychosocial outcomes than their Caucasian counterparts. Hispanic patients with CF have been shown to have poorer pulmonary function than Caucasian patients

(McGarry et al., 2017). African American and Hispanic CF patients have also been shown to experience poorer emotional health and social functioning than their Caucasian counterparts, even after controlling for SES and disease severity (Quittner et al., 2010). As a whole, minority status appears to be an important factor in disease outcome and experience, and therefore in treatment adherence.

PSYCHOSOCIAL SEQUELAE AND BARRIERS

The overall psychosocial strain strain of having CF is a notable barrier to treatment adherence, particularly during the developmental stage of adolescence. Time-consuming treatments may take up time that would otherwise be dedicated to socializing with peers in and outside school (Sawicki et al., 2014). Increased health consequences for developmentally-appropriate experimentation with risky behaviors in adolescence, such as smoking or drinking, may also make adolescents with CF feel socially isolated or unfairly penalized.

Higher levels of social support have been associated with better mental health, fewer physical symptoms, and less perceived treatment burden in adults with CF (Flewelling et al., 2019). Importantly, increased social support was not associated with increased treatment adherence or more adaptive beliefs about treatments, but was nonetheless associated with decreased perceptions of treatment burden and better mental health. As such, social support may alleviate some of the negative mental health outcomes from chronic illness. However, kids and adolescents with CF cannot make physical contact with each other, or even be in the same room. Breathing in proximity to each other could result in a deadly cross-infection of lung bacteria (Saiman & Siegel, 2003). As such, youth with CF can't access the same network of social support from peers as youth with other chronic illnesses outside of remote or electronic communication. In the absence of such an

important way to normalize kids' differences from healthy peers, youth with CF may feel more socially isolated and have more difficulty talking about their diagnosis with healthy peers than those with other chronic illnesses.

Similarly, negative mental health outcomes such as anxiety and depression may be more prevalent in kids and adolescents with CF than in the general population, and have been associated with poorer treatment adherence. A study of adolescents and adults with CF across nine countries found elevated anxiety levels compared to community samples, and more anxious patients were more likely to report experiencing chest symptoms (Quittner et al., 2014). The same study found that 10% of adolescents and 19% of adults had elevated symptoms of depression. Another study of kids and adolescents with CF found that 29% of patients had elevated symptoms of depression and that those with elevated scores had worse adherence to ACT (Smith et al., 2010). Depression has also been associated with declining lung function in people with CF (Fidika et al., 2014) as well as lower quality of life (Yohannes et al., 2012). As such, it is important to consider the psychosocial strain of dealing with a chronic medical illness such as CF in order to increase health-promoting behaviors and improve physical health outcomes.

Coping Skills

Given the various sources of distress caused by having a chronic illness such as CF, the ability to cope with distress is an important factor in overall health outcomes. The skills used to cope with various chronic illness are often divided into those that represent primary control, where efforts focus on changing the external source of distress, and secondary control, in which coping focuses on changing the internal appraisal or experience of the stressor (Rothbaum et al., 1982; Rudolph et al., 1995). Primary coping might involve using organization techniques to break down a complicated, multi-step treatment, using a

calendar to schedule recurring treatments or appointments, or seeking support to deal with the stressor. Using positive distraction, mindfulness, or reframing the stressor are types of secondary coping. By contrast, disengagement coping occurs when people do not attempt to use coping skills in response to a stressor. Those who favor disengagement coping may lessen their distress by avoiding the stressor. They may avoid talking or thinking about their illness. They may also avoid external, physical reminders of their illness by neglecting to monitor important health outcomes, missing medical appointments, or putting off treatments.

While no amount of primary control coping can cure an incurable illness, individuals with chronic illness may use it to motivate themselves to engage in health-promoting behaviors such as treatment adherence. Secondary coping may be more useful in adapting oneself to the realities of their chronic illness or treatment side effects that they can't change. Indeed, using primary coping has been associated with various desirable outcomes in people with diabetes, such as better quality of life and illness-related markers of health (Compas et al., 2012; Jaser & White, 2011). By contrast, more disengagement coping has been associated with worse illness-related health outcomes in diabetics (Graue et al., 2004).

People with CF have demonstrated roughly similar patterns of coping and outcomes (Abbott, 2003; Abbott et al., 2001). Adherent adults with CF were shown to use more secondary coping than nonadherent patients (Abbott et al., 2001). The use of social support has been associated with better psychosocial and physical health, health-related quality of life, and lower experience of treatment burden in adults with CF (Flewelling et al., 2019). By contrast, patients with poorer adherence to ACT and enzymes have been shown to use more disengagement coping than patients with higher adherence (Abbott et al., 2001). Using more disengagement coping has been associated with worse health-related quality

of life (Abbott et al., 2008). As such, coping skills can be an important conduit through which illness-related experiences are processed and appraised.

Illness Experiences

While physical health outcomes are often thought of as the direct result of treatment adherence behaviors, there is evidence that both negative and positive disease outcomes influence patients' appraisals of their illness, and that that perception influences their future adherence behaviors. For example, a patient who experiences chronic pulmonary exacerbations despite having maintained high treatment adherence since their last doctor's visit might begin to view their illness as less controllable, or the treatments as less effective. In line with Bandura's Social Cognitive Theory (1997, 1998), those beliefs might influence whether they take the time to engage in their treatments. Indeed, research has shown that adults with CF who had a pulmonary exacerbation within the past year regarded their illness as significantly more confusing and less controllable by treatments than those who had not (Sawicki et al., 2011). Greater variability in pulmonary function has also been shown to predict worse future treatment adherence behaviors for adults with CF (H. White et al., 2017). As such, it may be important for clinicians to consider the impact that past negative medical experiences have had on the beliefs people form about their illness, treatments, and ability to affect change in their illness. Helping patients reframe those experiences and beliefs may be an important intervention target.

Beliefs About Treatments

Past negative illness experiences can change people's beliefs about their illnesses, as well as the treatments they're prescribed for those illnesses. Naturally, patients' beliefs

about their treatments affect their adherence to those treatments, and as a result, health outcomes.

Necessity and Efficacy

Beliefs about whether a treatment is necessary may hinge upon subjective appraisal of lived experience. People with chronic illness who believe that their treatment is unable to cure or control their symptoms may be less likely to consider it a valuable part of their medication regime (Bucks et al., 2009; Law et al., 2014; Nouwen et al., 2009). Similarly, adherence tends to be better for medications that alleviate an existing discomfort, rather than those that must be taken proactively. Indeed, while kids generally have better adherence than adolescents, one study found that adolescents took a significantly greater proportion of mucus thinners (Dornase Alfa) than kids, as the medication relieved symptoms in real time (Zindani et al., 2006).

A study of adolescents with CF found that their self-reported adherence to ACT and antibiotics was significantly correlated with how necessary they believed the ACT was for them personally (Bucks et al., 2009). Further, beliefs about the necessity of medications predicted adherence over and above age in hierarchical linear regression. Belief in how effectively treatments could control symptoms was also a significant predictor of antibiotic use. Another study found that more consistent adherence to ACT in young adults was significantly associated with stronger belief in the necessity of the treatment and lower concern about side-effects (Sherman et al., 2019). Similarly, the belief that ACT could not ameliorate symptoms predicted significant variance in adherence to ACT in adults with CF (Myers & Horn, 2006). More negative beliefs about medications have also been shown to mediate the relationship between depressive symptoms and poor adherence to pulmonary medications (Hilliard et al., 2015).

Beliefs about the necessity of treatments affect adherence behaviors in other chronic illnesses as well. A meta-analysis of 94 studies assessing beliefs about treatments for various illnesses found that adherence behaviors were significantly related to stronger beliefs in the necessity of treatment and fewer beliefs about the harms of treatments (Horne et al., 2013). Similarly, a path analysis of factors predicting medical and mental health outcomes for adults with irritable bowel syndrome found that stronger beliefs about whether medications could cure or control their illness predicted health satisfaction directly, and indirectly by contributing to more active coping (Rutter & Rutter, 2002). In other words, those who believed their symptoms could be changed by medications took steps to do so. In the same study weaker beliefs about the efficacy of treatments predicted depression through their association with behavioral disengagement coping. Similarly, a path analysis of how beliefs about illness predict health outcomes for kids with chronic fatigue found that weaker trust in treatment control, more illness-related negative emotions, and greater perceived understanding of the disease led to lower quality of life through greater perseveration on symptoms (Gray & Rutter, 2007).

These beliefs appear to be malleable to intervention. One study of adults with chronic cardiovascular disease or type 2 diabetes created a series of tailored adherence interventions based on participants' self-reported barriers to treatment adherence (Nguyen et al., 2016). One of the interventions targeted beliefs about the necessity of treatment relative to concerns they had about the potential harms of treatments at baseline. Participants in the intervention that focused on changing beliefs about treatments had significantly greater beliefs in the necessity of treatment 3 and 6 months after the intervention. The interventions that focused on other barriers like forgetting to take medication did not result in substantial changes in beliefs about treatment necessity or concerns (Nguyen et al., 2016). Another intervention for people with asthma that tailored

text-messages based on beliefs about treatment necessity found that the intervention significantly increased their necessity beliefs and treatment adherence (Petrie et al., 2012). As such, beliefs about the necessity of treatment appear to be an important intervention target that can be a robust catalyst of behavior change.

Concerns and Harms

In addition to beliefs about the efficacy or necessity of treatments, beliefs about the potential harms or side-effects of treatments may also have direct behavioral implications and can prevent treatment adherence. Worries about the harms or negative side effects of treatment have significantly related to ACT adherence behaviors for adults with CF (Myers & Horn, 2006; Sherman et al., 2019). Parents of kids with high adherence to ACT have also been shown to have significantly higher beliefs about the necessity of treatment and significantly lower worries about potential harms than parents of low-adhering kids (Goodfellow et al., 2015). These findings indicate that patients' beliefs and concerns about treatments may be an important intervention target to improve adherence.

Beliefs About Agency and Illness

People's adherence is affected by their beliefs not only regarding treatments, but also regarding their illness itself. Individuals dealing with chronic illnesses have varying beliefs about how treatable their illness is, whether prescribed medications are necessary or sufficient to treat their illness, and about their own ability to manage their illness with those treatments (Bucks et al., 2009). Some such beliefs may be the result of misunderstanding information previously given by clinicians and may therefore be ameliorated by education. However, others may be influenced by selective memories of past negative health experiences (Sawicki et al., 2011), perceptions of invincibility or

exceptionalism specific to adolescence (Bucks et al., 2009; Suris et al., 2004), or negative beliefs about their own abilities to control their illness (Nouwen et al., 2009).

Beliefs about one's illness and treatments may be due to content knowledge about each. As such, some interventions have employed patient education initiatives to fill this deficit (Nguyen et al., 2016). However, many such programs have found that mere education about illnesses or treatments is necessary, but not sufficient for good treatment adherence or health outcomes (Bishay & Sawicki, 2016; Goldbeck et al., 2014; Savage et al., 2014; Tahmasebi et al., 2015). These interventions appear to be most helpful for the least adherent or patients burdened by low SES. Beyond those variables, having knowledge about treatments or illness may fail to reliably predict adherence behaviors and health outcomes because individuals may interpret that knowledge via their personal beliefs. In particular, people's adherence behaviors may be influenced by whether they believe their actions will be necessary or sufficient to affect change in their illness (Bucks et al., 2009; Sherman et al., 2019), or whether their illness is serious enough to warrant performing strenuous or aversive treatments.

Higher levels of self-reported personal control have been associated with better perceived physical health and social and emotional wellbeing in adults with CF, controlling for illness severity and CF-related markers of health (Sawicki et al., 2011). Personal control has also been associated with better adherence to diet restrictions and more positive health outcomes in people with diabetes (Broadbent et al., 2006, 2011; Wisting et al., 2016). The amount of personal control people believe they have over their illness may be an important and malleable intervention target for individuals with various chronic illnesses (Broadbent et al., 2006, 2011; Duff & Latchford, 2013). Interventions that target it and related constructs have been associated with improvements in treatment adherence and other physical and mental health outcomes (Keogh et al., 2011).

Similarly, self-efficacy is an important determinant of whether people believe they have the ability to affect change in their illness (Bandura, 1986, 1998). Higher treatment-related self-efficacy has been shown to predict more consistent adherence to ACT in people with CF (Sherman et al., 2019). However, constructs like personal control and self-efficacy have not been consistently associated with treatment adherence (Faint et al., 2017; Law et al., 2014). Instead, these constructs may affect adherence indirectly by influencing patients' ability to cope with their illness. One study found that children with CF's beliefs about their treatment self-efficacy and experience of illness burden accounted for significant variance in emotional adjustment to their disease, controlling for maternal education, child age, and disease severity (Thompson et al., 1998). Self-efficacy has also been shown to mediate the relationship between depressive symptoms and adherence to pulmonary therapies (Hilliard et al., 2015).

Overall, constructs that describe agency in disease outcomes relate to people's beliefs about their own competency relative to their beliefs about the difficulty of controlling their illness with the treatments available to them. As such, their adherence behaviors may reflect some interaction between their beliefs about their treatments, their illness, and themselves (Bandura, 1998). Repeatedly treating a chronic illness, often with varying results, presents myriad opportunities to reinforce or challenge those beliefs. People experience varying degrees of efficacy in treatment completion and symptom control, and sometimes experience that the former has little bearing on the latter despite their best efforts. Consistently being unable to lessen painful or debilitating symptoms that often limit participation in preferred activities can make one's illness feel as though it subsumes every aspect of one's life (Oris et al., 2016).

Chronic illnesses like CF can affect what aspects of life adolescents have access to, their peer relationships, and their beliefs about themselves. As such, it can sometimes be

difficult for them to know where their chronic illness ends and their own identity begins. However, given that adolescence is a vital time to establish identity, it is important that their illness can exist within their identity in a healthy way (Erikson, 1968; Oris et al., 2016).

Illness Identity

The degree to which people see their illness as a part of their identity, and the manner in which they do, can affect their illness-related behaviors and overall quality of life. This idea has been described through various constructs and terms, such as "Illness Self-Concept" (Morea et al., 2008), "Illness Perception" (Broadbent et al., 2006; Moss-Morris et al., 2002; Weinman et al., 1996), "Illness Cognition"(Evers et al., 2001), and, simply, "Illness Identity" (Oris et al., 2016). The construct of illness identity (Oris et al., 2016) and the corresponding Illness Identity Questionnaire (IIQ) emerged from Erik Erikson's (1968) work on identity development.

According to Erikson's model, identity develops in stages that correspond to unique, increasingly complex opportunities and demands that emerge as people move from infancy to adulthood and beyond. Each developmental stage includes age-appropriate milestones for physical and psychosocial development. Identity is formed, in large part, by the beliefs that people form about themselves and the world during these experiences and opportunities for growth. These beliefs are often based on their appraisal of how they have met the increasingly demanding tasks of each subsequent life stage. For example, the maturation of babies' physical abilities creates new opportunities to experience agency as they begin to interact more with their environment. Erikson's theory posits that babies either develop a sense of healthy autonomy or come to believe they have little personal

control based on whether they have the ability to experience agency and competency by affecting change in their environment.

Adolescence is a particularly important stage in identity development as it combines significant physical changes with substantial changes in opportunities, responsibilities, and consequences. Rapid cognitive development during adolescence creates new opportunities to experience independence from caretakers while physical maturation creates an incentive to explore new aspects of identity through peer relationships. Much of identity in adolescence is formed through peer appraisal, which makes adolescents' ability to assert themselves as individuals an even more salient aspect of identity development.

However, adolescents who experience chronic illness, or other life-disrupting events, may not be able engage in important aspects of identity formation for their life stage. Complicated treatment regimes often preclude full independence as they require parental intervention or assistance. Adolescents who are not given the opportunity to experience sufficient independence in managing treatments are more likely to feel that their illness subsumes their identity entirely (Raymaekers et al., 2020). Similarly, lengthy treatments that require time, space, or equipment inevitably interfere with time that could be spent exploring one's identity with peers (Sawicki et al., 2014). As such, chronic illness naturally creates barriers to the typical activities of identity development in adolescence. That said, having a chronic illness does not preclude the healthy formation of identity in adolescence. Instead, it simply requires that the reality of having a chronic illness is integrated as an aspect of the emerging adolescent identity.

Incorporating chronic illness into one's identity is not merely a conceptual or spiritual matter. Instead, the degree to which people see their chronic illness as part of their identity may affect how willing they are to alter their lifestyles to accommodate their

illness, and how doing so might affect their mental health (Oris et al., 2016). Many researchers in the field of chronic disease have sought to represent the construct of how people's illnesses affect or interact with their identities. In particular, the Illness Identity Questionnaire (IIQ; Oris et al., 2016) represents 4 distinct dimensions of illness identification, Rejection, Acceptance, Engulfment, and Enrichment. Each dimension represents aspects of illness identification that are hypothesized to relate to specific psychosocial, behavioral, and resulting medical outcomes.

Those who wish to Reject the idea that their illness is part of their identity are less likely to take the time to schedule treatments into their day. Instead, they may prioritize more identity-affirming activities, such as spending time with friends. These competing identities may be even more opposed during adolescence, a time when establishing one's identity is of paramount importance. Those who attempt to deny that illness is part of their identity may choose to engage with peers to the detriment of necessary developmental and illness-related activities, and yet still use lower levels of peer support to cope with their chronic illness (Raymaekers et al., 2020). Although they sacrifice treatments to spend more time with peers, they may avoid the topic of illness entirely and therefore miss out on opportunities for social support. People who feel they can't or won't accept their illness as part of their lives may not see their illness as something that will last their entire lives, or whose course could affect their future (Bucks et al., 2009). This is an especially pressing concern for adolescents who may already be less likely to consider the future ramifications of their current actions, either due to adolescent beliefs about invincibility or an acute awareness of an unavoidably foreshortened lifespan due to CF (Sawicki et al., 2014).

Rejection is hypothesized to result in more avoidant coping and poorer long-term mental health outcomes and treatment adherence (Oris et al., 2016). Those who reject their illness as part of their identity may engage in more disengagement coping, avoid thinking

or talking about their illness, and may avoid or put off treatments. The Rejection dimension has been associated with poorer treatment adherence in diabetic adolescents (Oris et al., 2016). While the avoidance may reduce their stress in the short term, Rejection results in poorer mental and physical health outcomes in the long term. For young adults with epilepsy, higher Rejection was associated with the perception of more negative side-effects from medication, more worry about seizures, and worse health-related quality of life and emotional well-being (Luyckx et al., 2018). Rejection has been associated with greater illness-related distress, symptoms of depression, illness symptoms, and lower life-satisfaction (K. M. Ross, 2018a).

On the other hand, healthy identification with one's illness could be a protective factor both for adherence behaviors and psychosocial outcomes. Illness Acceptance describes whether one can or will fit their illness into their identity and life. Greater illness Acceptance is hypothesized to relate to more active coping and result in better mental and physical health outcomes and higher treatment adherence. If someone is able to accept that the time they spend managing symptoms and adhering to treatments is a normal part of their lives, they may experience less psychosocial burden from their illness and be more likely to integrate treatments into their daily life (Luyckx et al., 2018; Oris et al., 2016; K. M. Ross, 2018b). Indeed, Acceptance has been significantly positively associated with treatment adherence and life satisfaction (Oris et al., 2016). Accepting one's illness has also predicted a healthier balance between peer relationships and responsibilities, and greater use of emotional support from peers (Raymaekers et al., 2020).

Acceptance has been associated with higher health-related quality of life in young adults with epilepsy, even after controlling for seizure severity and beliefs about medication (Luyckx et al., 2018). Higher illness acceptance was significantly negatively associated with anxiety, depression, and perceptions of health impairment in Dutch

adolescents with CF (Casier et al., 2008). Acceptance has also been shown to negatively relate to symptoms of depression and self-reported illness-related problems and positively relate to life satisfaction (Luyckx et al., 2018; Ross, 2018). Illness acceptance may also be associated with the belief that one's illness can be controlled, and that individuals personally have the ability to do so.

The experience of competence in one's ability to manage a complex illness may positively affect self-concept, self-esteem, and mental health. Some people may place importance on the positive experiences they've had as a result of their illness, such as increased social support, social bonds, or a unique perspective on life (Oris et al., 2016, 2018). As such, some people who positively incorporate their complex illness into their identity may perceive secondary benefits by framing their experiences as personal victories, or by considering any positive experiences they may have had as a result of their illness. This phenomenon of identifying benefits related to a stressful experience such as chronic illness has been described as illness Enrichment (Oris et al., 2016) and has been linked to post-traumatic growth (Oris et al., 2018; Tedeschi & Calhoun, 2004).

Patients with high levels of Enrichment might frame illness-related challenges as opportunities for growth. They might feel proud of the challenges they have surmounted and would be hypothesized to have conceptions of higher personal control over their illness, lower levels of negative mental health symptoms, and higher levels of life satisfaction. Enrichment may be an important indication of one's resiliency and ability to use secondary coping skills in response to illness-related challenges and has been associated with higher satisfaction with life (Oris et al., 2016). In a sample of young adults with epilepsy, Enrichment related to higher health-related quality of life and higher emotional well-being in hierarchical regression and correlation analyses (Luyckx et al., 2018).

However, identification with one's illness may not serve an exclusively protective role. Instead, people who feel that their illness occupies a significant degree of their identity or self-concept may feel that their illness has taken over their lives. Engulfment can occur when illness is perhaps too integrated into one's identity, to the detriment of other dimensions of identity. This sense of being Engulfed by their illness may result in poor psychosocial outcomes such as depression or anxiety, which have been associated with poorer physical health outcomes (Fidika et al., 2014; Hilliard et al., 2015). The Engulfment dimension has been associated with more depressive symptoms, more self-reported illness-related problems, and lower satisfaction with life (Oris et al., 2016). For epileptic young adults, Engulfment was associated with more worry about seizures, worse health-related and overall quality of life, emotional well-being, energy level, cognitive function, and social functioning, after controlling for perceptions of medication side-effects and seizure severity (Luyckx et al., 2018).

Engulfment may also be associated with the experience of more uncontrolled illness-related health outcomes. High scores on the Engulfment scale predicted hospitalization and outpatient medical care a year later for adults with coronary heart disease after controlling for age, illness complexity, sex, and symptoms of anxiety and depression (Van Bulck et al., 2018). Engulfment may occur when variability in health outcomes leads patients to doubt the efficacy of medications, the controllability of their illness, or their self-efficacy in changing illness outcomes. This may be why Engulfment is more consistently associated with both mental health outcomes and quality of life than with treatment adherence behaviors.

Overall, while the IIQ is a relatively new measure for illness identity, it touches on many topics central to the literature on illness identity and related constructs for adolescents with various chronic disease, and CF specifically. The Acceptance dimension captures

sentiments that reflect primary and secondary coping abilities while Rejection reflects more disengagement coping. The Engulfment and Enrichment dimensions illustrate how illness experiences can shape beliefs about agency and self-efficacy, which in turn can affect mental health. On the whole, the IIQ may provide a useful, holistic picture of how chronic illness affects identity, and how the resulting beliefs and mental health outcomes can affect treatment adherence.

THE CURRENT STUDY PROPOSAL

This study builds upon existing research on the value of assessing patients' illness experiences and beliefs to better understand and change their current and future adherence behaviors and health outcomes. The study will collect longitudinal data from adolescents with Cystic Fibrosis in the Central Texas area at four time points: intake, 6 and 12 months post intake, and 6 months before intake using retrospective chart review. Analyses will assess the relationship between illness identification and beliefs about treatment (Beliefs), adherence behaviors, and pulmonary function. The current study will assess the following hypotheses.

Research Questions and Hypotheses

RESEARCH QUESTION 1A

Does past pulmonary function predict future treatment adherence behaviors for adolescents with CF?

HYPOTHESIS AND RATIONALE 1A

Uncontrolled illness outcomes, represented by poor mean pulmonary function during the period up to 6 months prior to intake will be a significant predictor of poor adherence up to 6 months after intake, controlling for pulmonary function up to 6 months post and relevant demographic factors. While much research understandably focuses on the causal relationship between treatment adherence and future illness outcomes, greater variability in pulmonary function has been shown to predict worse future treatment adherence behaviors (H. White et al., 2017). Research has shown that negative past health experiences such as fluctuating health affect patients' beliefs about how confusing or uncontrollable their illness is (Sawicki et al., 2011). Given that beliefs about illness have

been shown to relate to treatment adherence and resulting illness-related health outcomes (Bucks et al., 2009; Horne et al., 2013; Oris et al., 2016), there is reason to believe that there may be a direct pathway between past pulmonary function and future adherence behaviors.

RESEARCH QUESTION 1B

Does past pulmonary function predict future treatment adherence by affecting adolescents' illness identity and beliefs about treatments during the intermediate period?

HYPOTHESIS AND RATIONALE 1B

Illness identity and beliefs about treatment (Beliefs) at intake will mediate the relationship between pulmonary function 6 months prior to intake and adherence 6 months after intake, controlling for adherence and pulmonary function at intake, pulmonary function and Beliefs 6 months post, and relevant demographic factors. Given that past negative pulmonary outcomes have been shown to predict worse future treatment adherence behaviors for adults with CF (H. White et al., 2017), and that less adaptive beliefs about treatment have been associated with treatment adherence for adolescents with CF (Bucks et al., 2009), there is reason to suspect that Beliefs would mediate the relationship between past negative pulmonary outcomes and future adherence behaviors.

RESEARCH QUESTION 2A

Do the beliefs adolescents with CF have about their illness and treatments predict future physical health?

HYPOTHESIS AND RATIONALE 2A

Reporting fewer adaptive Beliefs at intake will be a significant predictor of poor pulmonary function 12 months after intake, controlling for pulmonary function at intake, beliefs 12 months post, and relevant demographic factors. Less adaptive Beliefs will include higher scores on the Rejection and Engulfment and lower scores on the Acceptance and Enrichment scales of the IIQ, and a higher Concerns score and lower Necessity score on the Beliefs about Medicines Questionnaire-Specific (BMQ-S; Goodfellow et al., 2015). Negative beliefs about the necessity and efficacy of treatments have been shown to result in poorer adherence behaviors in various illness populations, including adolescents with CF (Bucks et al., 2009; Nouwen et al., 2009). Less adaptive health beliefs have also been associated with poorer psychosocial outcomes such as anxiety and depression in various illness populations (Casier et al., 2008; Rutter & Rutter, 2002), and poor mental health has been associated with poorer adherence (Hilliard et al., 2015) and worsening lung outcomes in people with CF (Fidika et al., 2014). As such, there is reason to believe that Beliefs will have an observable direct effect on future pulmonary function.

RESEARCH QUESTION 2B

Do the beliefs that adolescents with CF have about their illness and treatments predict future physical health by affecting interim treatment adherence?

HYPOTHESIS AND RATIONALE 2B

Adherence 6 months after intake will mediate the relationship between Beliefs at intake and medical status 12 months after intake, controlling for pulmonary function and adherence at intake, pulmonary function and beliefs 6 months post, beliefs and adherence 12 months post, and relevant demographic factors. Beliefs about the necessity and efficacy of treatments have been associated with poorer treatment adherence (Bucks et al., 2009;

Nouwen et al., 2009). Health beliefs have also been successfully targeted through interventions which resulted in significant improvements to treatment adherence (Petrie et al., 2012). Given the association between treatment adherence and physical health as measured by pulmonary function (Eakin & Riekert, 2013), there is reason to believe that adherence behaviors may mediate the longitudinal relationship between Beliefs and future pulmonary function.

METHODS

Participants

To be included in the study, participants must be between the ages of 11 and 17 at intake with a primary medical diagnosis of CF and be prescribed chest physiotherapy as a form of airway clearance at least once per day. Participants will be included if they use a mechanical high-frequency chest wall oscillation (HFCWO) vest to aid in airway clearance, as the current study will use the vest's existing usage log to measure treatment adherence. Participants will not be limited by CF strains which vary in severity of illness and intensity of treatment, but instead by whether they are prescribed ACT. The inclusion of a prescription for ACT effectively filters out the less treatment-intensive strains, which renders the exclusion of certain strains obsolete.

Procedures

Participants will be recruited from the Cystic Fibrosis clinic at Dell Children's Medical Center (DCMC) in Austin Texas. DCMC was chosen because it services the majority of pediatric patients with CF in the Central Texas area, and is the only medical care facility for many CF patients who live in surrounding rural areas. As such, DCMC acts as a hub through which patients gain access to services, and through which the researcher may access adolescents with CF. Patients are encouraged to have four scheduled visits per year to monitor health outcomes and disease progression. Patients and their families will be approached during routine checkups at the CF clinic, told about the study, and offered to participate.

Patients and their caregivers who choose to participate will be informed about confidentiality and the limits thereof, further informed about the study and use of the data, and asked to sign informed consent documents to give the researcher access to their CF-

related medical history for the past 6 months through retrospective chart review, as well as the following 12 months. The researchers will use retrospective chart review of the CF clinic's records to determine each patient's demographic information, including date of birth, age of diagnosis, assigned gender, race/ethnicity, and parental SES. Retrospective chart review will also be used to determine CF strain type, lung transplantation status, and whether the patient has experienced a pulmonary exacerbation or CF-related hospitalization in the previous 6 months. Illness-related health, defined by average pulmonary function measured during the past 6 months, will also be determined through retrospective chart review. On the day of intake, patients will fill out initial measures through Redcap, a secure and HIPAA-compliant electronic application used to distribute, collect, and store patient survey responses and data. Beliefs measures will be collected at intake and at 6 and 12 months post intake. Adherence for each timepoint will be calculated as a percentage by comparing the duration of vest use to the amount of time prescribed in each patient's chart. Illness Outcomes will also be collected at the 6 and 12 month point through chart review. Figure 1 provides a visual guide to what data are collected at each timepoint.

Measures

DEMOGRAPHICS

Participant demographics will be collected through retrospective chart review. Demographics will include age at intake, age of diagnosis, parental socio-economic status (SES), assigned gender, race/ethnicity, cystic fibrosis strain, and medical comorbidities. Age will be controlled for in all analyses due to its observed association with Beliefs and treatment adherence (Everhart et al., 2014; Smith et al., 2010). SES will be controlled for

due to its association with ACT adherence (Smith et al., 2010) and race/ethnicity will be controlled for due to its association with pulmonary health (McGarry et al., 2017).

ADHERENCE

Adherence to ACT will be measured via the usage log automatically collected by patients' mechanical high-frequency chest wall oscillation (HFCWO) vest, in the style of Oates et al. (2015). Adherence will be calculated as a percentage of time spent using the vest divided by the amount of time patients were prescribed to use the vest.

PULMONARY FUNCTION

Pulmonary function collected during each visit will be represented by the percent of forced expiratory volume a patient can produce in 1 second (FEV1), relative to a healthy peer.

BELIEFS

Beliefs about Medicines Questionnaire-Specific (BMQ-S; Goodfellow et al., 2015; Horne et al., 1999). The current study will use a version of the BMQ-S that has been adapted by Goodfellow and colleagues (2015) to assess how people with CF view CPT. The adapted scale includes 14 items and remains a clinically useful measure. The original BMQ-S contains two 5-item scales, Necessity and Concerns (Horne et al., 1999). Answers are given on a 5-point Likert scale ranging from Strongly Disagree to Strongly Agree, where higher scores indicate higher beliefs about necessity and concerns respectively. The questions from each scale are averaged to make up that scale's score. The Necessity scale measures beliefs about whether a medication is an integral part of maintaining health and the Concerns scale assesses beliefs about potential harms or side-effects the medication

could cause. Both original scales were initially normed on a sample representing six different chronic illnesses (Horne et al., 1999).

The original Necessity scale had internal consistency ranging from acceptable to good for all but one of the illness populations normed (Renal, $\alpha = 0.55$) . Average test-retest reliability for the Necessity scale was 0.77. The Necessity scale demonstrated construct validity through its positive association with scales measuring beliefs about the necessity of prescribed medications (Horne et al., 1999). Similarly, in people with chronic asthma, higher scores on the Necessity scale were associated with stronger beliefs that treatments can control their illness and greater concern about the illness itself (Broadbent et al., 2006). The original Concerns scale had internal consistency ranging from acceptable to good for all but two illness populations (Psychiatric, $\alpha = 0.63$; General medical, $\alpha = 0.65$) and test-retest reliability of 0.76. The Concerns scale's construct validity was shown through its association with items and scales measuring distrust of medications, desire for more information about medications, and increased sensitivity to their effects (Horne et al., 1999). Discriminant validity for both scales has been demonstrated through their ability to differentiate between different illness populations in which medication necessity and concerns are objectively different. The scales' ability to predict related outcomes, such as adherence behaviors, has been repeatedly demonstrated. A 2013 meta analysis of 94 studies found that higher Necessity and lower Concerns scores were significantly associated with higher treatment adherence for various chronic illnesses (Horne et al., 2013).

The BMQ-S was first adapted for a CF population by Bucks and colleagues (2009) to assess beliefs about ACT, enzyme therapies, and inhaled medications, but was further adapted by Goodfellow and colleagues (2015) to enhance the ACT scale's construct validity and predictive ability. Goodfellow et al. adapted the measure by adding four questions to the Concerns subscale to better match actual concerns that kids with CF and

their parents had raised about ACT during qualitative interviews, resulting in a total of 9 items. The additional questions assess whether ACT treatments make kids feel embarrassed or physically worse, and about negative side-effects from the treatments. Goodfellow and colleagues did not further amend the 5 questions on the Necessity scale for ACT (Goodfellow et al., 2015). Both scales were validated in a sample of 10 kids with CF and their parents and the internal consistency for both ranged from 0.75-0.90. To score the measure the authors summed each participant's scores on each scale and transformed them to a scale from 0-100 to account for the different number of questions on each scale. Scores will be coded such that higher scores on the Necessity scale and lower scores on the Concerns scale will indicate a more adaptive response. The current study will transform raw scores to z-scores to simplify analyses.

Illness Identity Questionnaire (IIQ; Oris et al., 2016). The Illness Identity Questionnaire (IIQ) was developed to explore how the degree to which people integrate their illness into their lives, and their identities, affects illness-related behaviors such as treatment adherence (Oris et al., 2016). The IIQ's 25 items assess cognitions that describe various manners of illness identification. The IIQ was initially validated in a sample of Dutch-speaking Belgians aged 14-25 with type 1 diabetes. It was found to have a clear 4-factor structure, resulting in dimensions of Acceptance, Rejection, Engulfment, and Enrichment (Oris et al., 2016). The Acceptance dimension describes whether people accept their illness as a part of their life, while Rejection describes whether people refuse to see their illness as part of their lives. The Engulfment dimension measures whether people feel every aspect of their lives are taken up by their illness while the Enrichment scale describes whether people can view illness-related challenges as opportunities for personal growth. The scales appear to measure related but discrete constructs. The Acceptance dimension has been significantly negatively correlated with the Engulfment and Rejection dimensions

and positively correlated with the Enrichment dimension in multiple illness populations (Luyckx et al., 2018; Oris et al., 2016). Similarly, the Engulfment dimension has been shown to be significantly negatively correlated with the Enrichment dimension and positively correlated with the Rejection dimension. Content validity of the scales was ensured through the nature of the development; the IIQ's items were based on existing, validated scales of illness identity and thematically related concepts.

The IIQ was later validated in an adult sample of Belgian adults with either congenital heart disease (n = 337) or multisystem connective tissue disorders (n = 241; Oris et al., 2018), and 121 Norweigan young adults with refractory epilepsy (Luyckx et al., 2018). The English version of the IIQ was validated in a national sample of 282 American adults with type 2 diabetes and was found to maintain the four-factor structure (Ross, 2018). Across illness populations and languages, internal consistency has ranged from acceptable to excellent for all scales. Cronbach's alpha values have ranged from 0.82 to 0.86 for Acceptance, 0.75 to 0.84 for Rejection, 0.90 to 0.92 for Engulfment, and from 0.90 to 0.95 for Enrichment (Luyckx et al., 2018; Oris et al., 2016, 2018; Ross, 2018). The test-retest reliability of the IIQ was demonstrated in a Dutch-speaking sample of 545 adolescents and young adults with type 1 diabetes when participants were given the IIQ at baseline, one year later, and two years later (Raymaekers et al., 2020). Each scale was significantly correlated with itself across every time point comparison.

The IIQ's test-retest reliability supports the idea that it measures a lasting trait developed through the integration of experiences and beliefs, rather than a fleeting state. The criterion validity of the IIQ scales has been established by comparing each scale to conceptually related outcomes (see above review). The measure's 25 items include 6 items in the Acceptance scale, 5 in the Rejection, 8 in Engulfment, and 8 in Enrichment. For each item participants indicate how much they agree with each statement on a Likert scale from

1 (strongly disagree) to 5 (strongly agree). The measure is scored by calculating the average for each scale, resulting in 4 dimension scores. Scores will be coded such that higher scores on Acceptance and Enrichment and lower scores on Engulfment and Rejection will indicate a more adaptive response. Scores will be transformed to z-scores to simplify analyses.

ANALYSES

The psych package from R will be used for all analyses (Revelle, 2013; R Core Team, 2018). Missing data will be dealt with using pairwise deletion within each hypothesis. The study includes two hypotheses that each use data from three timepoints. The latter hypothesis' last timepoint is 12 months post intake, when the study is likely to see the most attrition. As such, using pairwise deletion will allow participants who miss the last timepoint to be included in the former hypothesis.

The variable block that consists of the four IIQ scales and two BMQ-S scales will be referred to as "Beliefs" in subsequent descriptions of analyses. The Beliefs measures in this study are represented by a block of six quasi-interval scales that are each created by averaging the various ordinal items within that scale. The variables in this block serve as the predictor for hypothesis 1 and as the mediator for hypothesis 2. However, analyses that use the General Linear Model to understand relationships between data make assumptions that preclude the use of ordinal data as predictors. The author will perform analyses to determine whether the ordinal-level independent variables (IV's) can be treated as continuous and used in subsequent regression analyses. The raw ordinal data, which will be collected and stored as numeric responses, will be transformed into new variables where they are treated as ordinal. This transforms each variable using orthogonal polynomial coding and automatically sets their contrasts to that of an 11-level ordered factor. Doing so allows ordinal data to be entered as predictors or covariates in regression models, and returns data on the ordinal variable's linear, quadratic, and cubic prediction of a continuous dependent variable (DV). These regression outputs will determine whether the ordinal variable's prediction of the continuous dependent variable can be fit to a linear model (Chambers & Hastie, 1992), rather than a quadratic or cubic one. If so, the relationship

between the ordinal predictor and continuous dependent variable can be described as linear and monotonic.

To further test the appropriateness of treating the ordinal predictors as continuous numeric data, ordinal predictors with a demonstrated monotonic relationship to the dependent variable will be transformed into new categorical variables and compared to models where the predictor is treated as continuous numeric data using Likelihood Ratio tests. If the models using categorical and continuous versions of the same variable do not differ significantly in their ability to fit the data, and if the effects of the IV on the DV appear to increase in roughly consistent increments, the predictor will be treated as continuous numeric data in subsequent regression analyses for simplicity. If the model using the continuous version of the predictor provides a significantly poorer fit the variable will be dropped as a predictor from subsequent regression analyses. The author will operate under the assumption that any quasi-interval variables can be treated as numeric and continuous for the remainder of this proposal.

Prior to analyses, all interval and ratio variables will be converted to z-scores to ease interpretation and reduce the likelihood of multicollinearity of predictors. The regression for each hypothesis will be tested for assumption violations. For variables measured on an ordinal scale, linearity will be assessed through the aforementioned comparison of regression models. The linearity of data measured on a continuous scale will be assessed through visual inspection of scatterplots. The normality of the distribution of residuals will be checked through visual inspection of histograms or a goodness of fit test. Homoskedasticity will be assessed through visual inspection of scatterplots comparing the residuals to predicted values. Multicollinearity between independent variables and covariates will be assessed through correlations between the various predictors.

The mediate function from the psych package in R will be used for mediation regression analyses (Revelle, 2013; R Core Team, 2018). The mediate function defines and calculates mediation according to the method outlined by Hayes (2017; 2018). Hayes' procedure specifies that mediation occurs when the indirect effect (ab) of the predictor (X) on the dependent variable (Y) through the mediator (M) is significantly different from zero at the population level. Hayes recommends testing this hypothesis through bootstrapping, which treats the sample's indirect effect as a point estimate of that of the population. This procedure generates thousands of new random samples based directly on the values of the original sample. Bootstrapping repeatedly samples the original values, allowing for repeated values, to generate a distribution of samples that more closely approximate the population. The thousands of generated samples are used to estimate the distribution of indirect effect values expected at the population level. Using typical confidence intervals, if the 95% confidence interval for the population's estimated indirect effects does not contain zero, it is standard practice to consider the effect significantly different from zero and grounds to reject the null hypothesis (Hayes, 2017).

Unlike other procedures for establishing mediation (Baron & Kenny, 1986), Hayes' method does not require a significant total effect of X on Y to begin mediation testing (Hayes, 2018). While Hayes' method does estimate the total and direct effects to better understand the overall mediation model, mediation can exist independent of their significance. As such, the author will test hypotheses 1b and 2b regardless of whether hypotheses 1a and 2a establish significant direct effects. The total effect of X on Y (c) is the change in Y for one unit of change in X, not holding M constant. It can be estimated by regressing Y on X, without including M in the formula, and is equal to the sum of the direct (c') and indirect effects (ab) of X on Y ($c = c' + ab$) when using Ordinary Least Squares Regression with continuous data (Hayes, 2017). The direct effect of X on Y (c')

is the change in the expected value of Y with one unit of difference on X, holding M constant. Regression formulas for this study in R syntax can be located in the appendix.

Power Analysis

The power analysis was conducted using the Monte Carlo Power Analysis For Indirect Effects calculator created to determine sample sizes needed to detect mediation for up to 2 parallel mediators (Schoemann et al., 2017). The calculator uses known or likely correlations between aspects of a mediation model to calculate the sample needed to detect the effect of ab. Given the large number of parallel mediators in Hypothesis 1b and the fact that existing effect size values for an adolescent CF population do not exist for many of these measures in the CF community, conservative composite values were generated based on available associations between the measures of interest for various chronic illness populations. The BMQ and BIPQ scales were used together to estimate possible relationships between all the mediators and the other variables in the model. Existing correlations between adherence behaviors and the the BMQ's Necessity scale ranged from 0.32 to 0.40, and -0.40 to -0.43 for the Concerns scale, so values of 0.35 and -0.40 were used respectively for estimates of the correlation between the mediators and Y, Adherence (Bucks et al., 2009; Robert Horne & Weinman, 2002; Sherman et al., 2019). Values of 0.30 and -0.30 respectively were used to form a more conservative estimate of the mediators' relationship to X, Pulmonary Function. The correlation between the two BMQ scales has ranged from -0.19 to 0.31 (Robert Horne et al., 1999; Robert Horne & Weinman, 2002), so a value of 0.30 was used to be conservative in the case of potential multicollinearity for the correlation between the mediators. Increased adherence to treatments has been shown to be correlated with an improvement in FEV (r = 0.53), so a more conservative value of 0.50 was used for the relationship between X and Y (Moore et al., 2013). Inputting these

values resulted in a necessary sample size of 90 to detect the indirect effect of the first mediator, and 86 to for that of the second. In order to account for roughly 25% attrition seen in studies with similar clinic settings, the current study will seek 120 participants to maintain at least 80% power to detect the least robust indirect effect.

SUMMARY AND IMPLICATIONS

The current study would advance the existing research on how examining beliefs about one's illness and treatments can affect health-promoting behaviors and physical health outcomes for adolescents with serious chronic illness. This study would also provide mental and physical health clinicians with a more holistic picture of why some adolescent patients with chronic illness have more difficulty adhering. In particular, it would allow clinicians to identify adolescents with more variable illness-related health as potentially at risk for future negative mental health outcomes and poor treatment adherence, and provide them with targeted areas of intervention. Similarly, it would allow clinicians to quickly identify internal barriers to treatment adherence for chronically non-adherent adolescents.

Regarding the first hypothesis, patients with poorer past pulmonary function would be expected to have less adaptive interim health beliefs, scoring lower on Acceptance, Enrichment, and treatment Necessity and higher on Engulfment, Rejection, and treatment Concerns, given past research in the field. Those individuals would also be expected to have lower future treatment adherence. This pattern of results would indicate that patients' interpretation of past medical experiences informed their feelings about their illness and their beliefs about the efficacy and potential dangers of their treatments, and that those beliefs affected their future treatment adherence. This information would allow clinicians to identify patients with poorer past health outcomes as potentially at risk for future poor adherence and negative mental health outcomes associated with Engulfment and Rejection such as anxiety or depression (Oris et al., 2016).

Having information about the type of maladaptive illness identification patients experienced would allow clinicians to address the specific types of coping skill deficits theoretically associated with each (Oris et al., 2016). Using measures that can quickly

identify malleable core beliefs affecting adherence behaviors would be a clinically useful tool to structure interventions. Knowing that patients' beliefs about the necessity or harms of treatments affected their adherence behaviors would allow clinicians to create individualized interventions for non-adherent clients, rather than provide them with general information they may have already received.

Regarding the second hypothesis, those with less adaptive health beliefs at intake would be expected to have poorer adherence 6 months post and lower pulmonary function 12 months post. This would demonstrate that a patients' health beliefs assessed at any given medical visit has the potential to affect their future physical health through their interim adherence behaviors. Such information would likely be a compelling case for integrating mental health interventions into regular medical visits for those with chronic illness.

One limitation of this study is the use of pairwise deletion to deal with missing data within each study hypothesis. Given the longitudinal nature of the study much of the missingness is likely to occur in the last time point (i.e. 12 months post intake) which is used only by the latter hypothesis. As such, it is possible that the groups included in each hypothesis could be different in ways that could indirectly relate to study constructs. For example, participants from families with limited resources or who live in rural areas may be less able to attend regular medical appointments. The additional stress of managing transportation to a medical appointment could increase patients' experience of feeling overwhelmed by their illness. Alternately, those with more uncontrolled illness outcomes or those who find their illness more confusing might be more motivated to attend medical appointments. Multiple imputation would likely be a more appropriate choice for missing data but it is beyond the scope of the current proposal.

This study's use of ordinally-derived data as a parallel predictors in regression could also be considered a weakness. Although the author included methods in the analyses

to test whether the data could be treated as numeric, this method is not a perfect solution for including ordinal variables in regression models as it assumes equal distances between levels of the variable, provided that the data can fit the DV in a linear fashion. Another method of transforming the ordinal data, such as penalized regression (Gertheiss & Tutz, 2009) or use of ordinal smoothing splines (Helwig, 2017), may create a better-fitting model of the data by imposing systematic restrictions on the magnitude of differences in the DV for varying levels of an ordinal predictor. Such methods do not make assumptions about the monotonicity of the relationship between the ordinal predictor and continuous dependent variable, which may allow for better fit of the data. Other researchers (Bürkner & Charpentier, 2018) have developed a method for treating ordinal predictors that models their relationship with the dependent variable as monotonic, but which does not assume equal distances between levels of the predictor on the dependent variable.

Alternatively, the author could seek out more advanced analyses such as Structural Equation Modeling (SEM). SEM could be used to treat the ordinal variables as observable markers of unmeasured latent variables that are normally distributed and have a monotonic relationship to the DV, regardless of whether the observed variable does. As such, the assumption of normality in SEM would not be violated. However, these methods are currently beyond the author's level of understanding.

Another limitation of this study is that it uses a convenience sample from one Cystic Fibrosis clinic in Central Texas, which may not be representative of the entire CF population. A future study could obtain stratified random samples from multiple clinics across the state, or ideally across the country. Using a larger, more representative sample would also address another limitation in how the current study defines what constitutes a negative past health experience. A future study with a larger sample could stratify participants based on whether they had experienced a pulmonary exacerbation in the last 6

months. This would provide a more robust assessment of how, and which types, of past health experiences affect health beliefs.

While constructs like illness identity provide an interesting way to conceptualize the system of beliefs that drive differential treatment adherence and mental health outcomes in chronic illness, they are only meaningful if they allow clinicians to identify and address these beliefs more accurately than related constructs. Future studies could build upon the current study by assessing how the construct of illness identification compares to theoretically related constructs like coping skills and treatment-related self-efficacy in its ability to predict future adherence behaviors and physical and mental health outcomes. The current literature on the IIQ, to the best knowledge of the author, has not yet compared its ability to predict health-promoting behaviors relative to the aforementioned constructs. However, the existing research indicates that the IIQ, combined with an assessment of beliefs about treatments, would provide clinicians with valuable, actionable information about how patients' appraisals of past experience affect adherence and mental and physical health outcomes.

APPENDIX

Table 1: Adherence Rates for Cystic Fibrosis Treatments

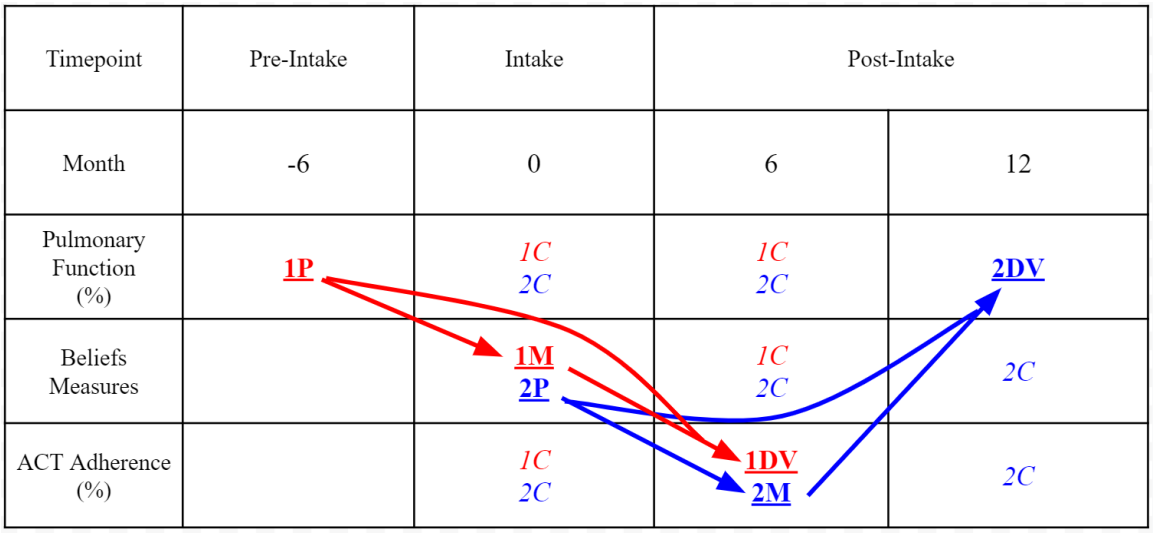
Affected System	Treatments	Adherence Rates (%)[a]
Gastrointestinal	PERT[b]	42.5[1]-49.4[2]
	Acid Blockers	67.0[3,i]
	Vitamins	33.7[1,ii]-67.7[4]
	Nutritional Supplements	50.0[5, iii]
Pulmonary	Inhaled Antibiotics	31.0-53.0[6,iv]
	Mucus Thinners	58.0-67.1[4]
	Corticosteroids	58.4[7,v]
	Bronchodilators	51.0[3,i]
	ACT[c]	60.0[8]
Genetic	CFTR[d] Modulator	61.0[9]

Note. [a]All figures represent mean adherence for patients with Cystic Fibrosis measured through electronic monitoring unless otherwise specified. [b] Pancreatic Enzyme Replacement Therapy. [c] Airway Clearance Therapy. [d] Cystic Fibrosis Transmembrane Conductance Regulator.

Data are from Modi et al. (2006)[1], Barker & Quittner (2016)[2], Rouzé et al. (2019)[3], Zindani et al. (2006)[4], Arias Llorente et al. (2008)[5], Cockburn et al. (1987)[6], (Milgrom et al., 1996)[7], (Oates et al., 2019)[8], and (Siracusa et al., 2015)[9].

[i] Adherence measured through Continuous Medication Availability (CMA) ratio. [ii] Pharmacy refill data. [iii] Self-reported adherence. [iv] Pill count. [v] Pediatric asthma population, median.

Figure 1: Temporal Overview of Mediation Analyses



Note. Description of data collected at each timepoint, as well as the timeline of data used in mediation analyses. The items in red describe the mediation model of hypothesis 1b, and the items in blue describe the mediation model of hypothesis 2b. 1P = Model 1b's predictor, 1M = Model 1b's mediator, 1DV = Model 1b's dependent variable, 1C = Model 1b's control variables. 2P = Model 2b's predictor, 2M = Model 2b's mediator, 2DV = Model 2b's dependent variable, 2C = Model 2b's control variables.

Table 2: Measurement Scale and Type

Measure	Subscale	Item Measurement	Item Level/range	Outcome Scale
FEV1	-	Percentage	0 - 100	Ratio
BMQ-S	*Necessity*	5 Ordinal Items	1 (*strongly disagree*) - 5 (*strongly agree*)	Quasi-Interval
	Concerns	9 Ordinal Items	1 (*strongly disagree*) - 5 (*strongly agree*)	Quasi-Interval
IIQ	*Rejection*	5 Ordinal Items	1 (*strongly disagree*) - 5 (*strongly agree*)	Quasi-Interval
	Acceptance	5 Ordinal Items	1 (*strongly disagree*) - 5 (*strongly agree*)	Quasi-Interval
	Engulfment	8 Ordinal Items	1 (*strongly disagree*) - 5 (*strongly agree*)	Quasi-Interval
	Enrichment	7 Ordinal Items	1 (*strongly disagree*) - 5 (*strongly agree*)	Quasi-Interval
Minutes of Actual Use / Prescribed Use	-	Percentage	0 - 100	Ratio
Age	-	Chart Review	-	Ratio
Race	-	Chart Review	-	Nominal
SES	-	Chart Review	-	Ordinal

Table 3: Components of Each Variable Block

Variable Block	Measure	Subscale
Pulmonary Function	FEV1	-
Beliefs	BMQ-S	*Necessity*
		Concerns
	IIQ	*Rejection*
		Acceptance
		Engulfment
		Enrichment
Adherence	Minutes of Actual vs. Prescribed Use	-
Demographics	Age	-
	SES	-
	Race/Ethnicity	-

Figure 2: Analyses Represented in R Syntax

Regression formulas are represented using the notation set by the *mediate* function in R: Y ~ X + (Mediator 1) + …. (Mediator k) +/- Covariate 1 +/- Covariate k. In R covariates can be included in the model using "+" or partialled out using "-". The variance explained by control variables will be partialled out in this study.

Hypothesis 1a:

Adh_6mopst ~ FEV_6mopre - FEV_6mopst - Beliefs_6mopst - Demographics

Hypothesis 1b:

Adh_6mopst ~ FEV_6mopre + (Beliefs_int) - FEV_int - Adh_int -
FEV_6mopst - Beliefs_6mopst - Demographics

Hypothesis 2a:

FEV_12mopst ~ Beliefs_int - FEV_int - Adh_int -
Beliefs_12mopst - Adh_12mopst - Demographics

Hypothesis 2b:

FEV_12mopst ~ Beliefs_int + (
FEV_6mopst) - FEV_int - Adh_int - FEV_6mopst - Beliefs_6mopst - Belie
fs_12mopst - Adh_12mopst - Demographics

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