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Acknowledgments

This would not have been possible without the support of my mentor, Dr. Beth Wildman. She has been supportive, encouraging, and has provided incredible guidance throughout my growth in graduate school. I could not be more thankful. I also must thank Dr. Bodas, who first pulled me into the wonderful world of sickle cell and whose passion has lit a fire and interest that will not soon be extinguished. The support of Dr. Grau, initially as a clinical supervisor and now on my committee has been invaluable. Dr. Updegraff has served as a great resource on this committee and throughout graduate school for growing my knowledge of social health psychology. And of course, I appreciate the willingness of Dr. James to join us and provide his expertise and time during the defense.

I also must acknowledge and profoundly thank Lisa Sidebotham and the staff of the Akron Children’s Hospital Sickle Cell Program. The quality of this research would have suffered without their help and intimate knowledge of the families that participated. Thanks and the utmost respect to the families that participated in our research. They always had the intent of bettering the state of knowledge for those adolescents with sickle cell to follow.

An incredible thank you to my lab mates – Rob, Heather, Nicole, Shana, Kara, and Jeannette. I’m so lucky to have a professional family that have also become such great friends and supports throughout this process. Finally, thank you to my family – Chandler, Mom, Dad, and Grandma – who taught me to dream big, be stubborn about my goals but flexible about my methods, and allowed me the grace to fail safely along the way. This is because of you.
CHAPTER 1

INTRODUCTION

Sickle Cell Disease (SCD) is a genetic, lifelong condition in which the body produces red blood cells that are sickled. The symptoms of SCD include pain crises due to blocked blood flow, acute chest syndrome, increased risk of infection, decreased red blood cells, and stroke (Centers for Disease Control, 2013; Roseff, 2009). SCD affects individuals with family lineage from Africa, South or Central America, Caribbean islands, Mediterranean countries, India, and Saudi Arabia. The life expectancy of individuals with SCD is approximately 50 years (Claster & Vichinsky, 2003), and mortality rates of older children and younger adolescents (aged 10-14) have not improved since 1983 (Yanni, Grosse, Yang, & Olney, 2009). If SCD is well managed, the patient can decrease the risk of early mortality and the number of complications related to the condition (Claster & Vichinsky, 2003). The regimen of care for individuals with SCD typically includes guarding against infection through prophylactic penicillin and regular vaccinations, as well as frequent monitoring to track incidence of stroke (Claster & Vichinsky, 2003). For some patients, their care also includes regular blood transfusions (Roseff, 2009). The only cure for SCD is bone marrow transplant, a risky and difficult procedure that often involves a sibling donor, but results in an event-free survival rate of greater than 90% (Bernaudin et al., 2007).

Due to the chronic and difficult to manage nature of SCD, non-adherence is a problem. Rates of adherence to disease management tasks range from 12% (Elliott, Morgan, Day, Mollerup, & Wang, 2001) to greater than 80% (Thornburg, Calatroni, Telen, & Kemper, 2010).
depending on the measure utilized. A recently published meta-analysis of medication adherence in pediatric SCD found rates ranging from 12% to 100% and attributed the range to varied assessment strategies (Loiselle et al., 2016). In that meta-analysis, adherence rates by objective methods ranged from 43% to 94.4%.

Non-adherence in adolescents with SCD leads to greater rates of hospitalization, infection (Ahmed, 2011), pain crises, acute chest syndrome, and an increased risk for early death (Platt et al., 1991). These symptoms can have secondary impact on daily functioning, such as sleep and school attendance (Schatz, Brown, Pascual, Hsu, & DeBaun, 2001; Shapiro et al., 1995). Nonadherence is a complicated, multi-faceted problem, and must be addressed through patient factors as well as members of the healthcare team that care for the patient daily, including parents (DiMatteo, Haskard-Zolnierek, & Martin, 2012). The current study examines parent and adolescent factors that contribute to adherence in adolescents with SCD and the associated health outcomes.

Due to the relative lack of racial diversity in SCD, it is important to address diversity of other demographic variables, particularly poverty status and socioeconomic status (SES). In studies of youth with SCD and their caregivers, family income varies, with up to 46% reporting under $20,000, and more than half (55%) living in neighborhoods with a poverty rate higher than 27.4% (Palermo, Riley, & Mitchell, 2008). The distress of poverty and complicated socioeconomic factors affect research in this population. In one study of youth with SCD, unstable home environments such as serial foster homes resulted in 15.8% of the potential sample being excluded from the study (Treadwell et al., 2005). Parent education is also relatively low in this population. Studies report between 44.9% and 75% of parents have no more than a high school education. Insurance type has also been a measure of SES, and a recent meta-
analysis of pediatric SCD reported the average percentage of participants with public insurance was 89% (Loiselle et al., 2016). Poverty status and SES both significantly impact youth with SCD. SES data (parent education and family income) significantly predicted the child or adolescent’s functional disability and physical and psychosocial health-related quality of life in a sample of youth with SCD (Palermo et al., 2008). Privately insured patients with SCD have higher adherence than those with public insurance (Teach, Lillis, & Grossi, 1998), and family income is positively associated with adherence ratings (Barakat, Smith-Whitley, & Ohene-Frempong, 2002). Youth with SCD must cope with having a chronic illness, racial minority status, as well as frequent poverty and SES concerns as exhibited by such studies.

Disease management in adolescents with chronic illnesses is more complex than for young children or adults. As they are developing, adolescents become more independent and assume more responsibility for their adherence compared to younger children (Andersen, 1995; Drotar & Ievers, 1994; Manne, Jacobsen, Gorfinkle, Gerstein, & Redd, 1993; McQuaid et al., 2001). Unfortunately, in other populations with chronic illness, such as asthma, adherence often decreases during this time of transition as parents expect adolescents to take more responsibility for their disease management, but the adolescent does not fulfill the expected responsibility (McQuaid, Kopel, Klein, & Fritz, 2003). Fewer studies have examined responsibility for disease management tasks in SCD, with one study finding parent and youth report of responsibility for disease management falling more to the parent’s role (Oliver-Carpenter, 2011). Parents’ inappropriate developmental expectations of independence in adherence during this time result in poor overall adherence for adolescents [for example, with asthma,(Walders, Drotar, & Kercsmar, 2000)] due to the mismatch in giving up responsibility (by the parents) and taking on the
responsibility (by the adolescent). A recent meta-analysis found that adherence is poorest in older adolescents with SCD (Loiselle et al., 2016).

Children and adolescents with SCD who share more responsibility for adherence with their parents have greater knowledge of their disease than youth who share fewer tasks, both in groups whose parents were mainly responsible and groups in which the adolescent was mainly responsible. Non-adherent children and adolescents with SCD are less likely to share adherence tasks with their caregivers (Treadwell & Weissman, 2001). Adolescent renal transplant candidates are more likely to adhere to their treatment and have fewer missed doses of medications when their parents are involved in responsibility of disease management (Zelikovsky, Schast, Palmer, & Meyers, 2008), highlighting the need for continued parental supervision during adolescence. In pediatric asthma, older age has been related to an increase in responsibility for asthma adherence (McQuaid et al., 2003). Additionally, cognitive maturity has been related to an increase in responsibility in adolescents with diabetes (Ingersoll, Orr, Herrold, & Golden, 1986). As a result of the complicated relationship of responsibility for adherence and the developing independence during adolescence, shared responsibility for SCD care has evidence as the best approach during adolescence (Treadwell & Weissman, 2001).

**Models for Studying Adherence in Chronic Illness**

Previous research has revealed that many factors contribute to adherence in SCD, and understanding of the best model for studying adherence in this population can be informed by models of adherence in chronic illness literature. While no models have been systematically tested in adolescents with SCD, researchers suggest applying disease-specific models to other chronic conditions in order to further adherence research (La Greca & Mackey, 2009).
childhood, Type 1 Diabetes is one pediatric chronic illness with significant research surrounding a complicated adherence regimen.

For pediatric diabetes, intervention for adherence must include the family system and not just the individual adolescent patient. When examining multiple studies on pediatric diabetes care, communication, problem solving skills, and supportive involvement from the family system affect adherence and care (Wysocki, Buckloh, & Greco, 2009). One model for understanding adherence in adolescents with diabetes suggests that the relationship between family variables (family functioning, parental guidance and control, parental warmth and caring, parental criticism and negativity, no responsibility for diabetes regimen) and disease control (metabolic control) is mediated by adherence (Lewin et al., 2006; Lewin et al., 2009).

In addition to utilizing diabetes-specific models of care, the Health Belief Model is useful for understanding adherence to disease management more broadly. The Health Belief Model [HBM; (Janz & Becker, 1984)] factors include perceived susceptibility, perceived severity, perceived benefits, perceived barriers, and cues to action. Perceived susceptibility includes an individual’s understanding or acceptance of his or her risk for negative health outcomes. Perceived susceptibility has been correlated positively or negatively with adherence to treatment, depending on the disease being studied (Rapoff, 2010). Perceived severity includes the intensity of negative consequences individuals believe may come from not following (or following) a recommended health behavior as well as the perceived severity of their disease diagnosis. A meta-analysis of studies utilizing the HBM to understand adult and pediatric patients’ adherence to treatment regimen found that greater perceived severity threat of their illness is associated with better adherence (DiMatteo, Haskard, & Williams, 2007). However, age moderated the results, as among the pediatric samples in the meta-analysis, there was a negative association
between objective measures of disease severity and adherence. Pediatric patients in poorer health (higher severity) had poorer adherence compared to pediatric patients in better health. There was some evidence that parent-rated disease severity was negatively related to adherence in patients with more serious diseases (e.g., diabetes) and positively related for those with less severe diseases (e.g., asthma). That is, for those with serious conditions and high parent-rated disease severity, adherence was poor; those with less serious condition and high parent-rated disease severity, adherence was better.

Perceived benefits are the positive outcomes an individual may expect to receive as a result of engaging in health behaviors. Perceived benefits to adherence were positively related to adherence in a study utilizing the HBM in adolescents with insulin dependent diabetes mellitus [IDDM; (Bond, Aiken, & Somerville, 1992)]. Benefits also interacted with a measure of perceived severity and perceived susceptibility to indicate that those with low severity and susceptibility and high benefits were the most adherent to their regimen. Perceived barriers include the impediments, difficulties, or other concerns an individual has to make it more difficult to engage in a health behavior or prevent him or her from engaging in a health behavior. This may include an analysis of the costs versus the benefits an individual calculates of engaging in a certain behavior related to adhering to treatment. Research has shown that perceived barriers are a strong predictor for health practices across a range of conditions, ages, and health behaviors, including mother’s adherence to regimens for managing her child’s conditions (Janz & Becker, 1984).

Finally, cues to action are reminders to engage in a health behavior. These are related to better adherence to a medical regimen for adolescents with diabetes (Bond et al., 1992). More recently, self-efficacy has been added as an important factor in the HBM (Strecher &
Rosenstock, 1997). In a meta-analysis of studies of adults utilizing the HBM to understand adherence, all elements of the model were related to better adherence, though specific importance of an element varied according to the study (Harrison, Mullen, & Green, 1992).

The HBM has been utilized to study adherence in youth with SCD in only one known study. In a study applying the model to parents of children (ages 6 to 60 months) with SCD, HBM factors accounted for about a third of the variance observed in adherence rates, with perceived burdens of picking up medication and forgetting being the most significant contributing factors (Elliott et al., 2001). The HBM has also been applied to adolescents with other chronic conditions, including chronic asthma (McQuaid et al., 2003). In this study, adherence was not related to youth (ages 8-16) knowledge, reasoning, or responsibility for adherence. In a study applying the model to children and adolescents (age 10-19) with IDDM (Bond et al., 1992), results showed that as age increased, adherence decreased and perceived threat increased. Cues to action (which, in this study, were conceptualized as a willingness to seek help when experiencing diabetic symptoms) were closely related to adherence to their treatment regimen. When adolescents perceived low benefits for cost, threat was related positively to adherence. When adolescents perceived high benefits for cost, threat was negatively related to adherence. This study indicates that, when studying adolescent adherence to prescribed treatments, threat is complicated and does not relate to adherence as expected or predicted by the HBM.

Utilizing the HBM is difficult in adolescence due to its reliance on only the understanding of the adolescent’s health beliefs, which are likely skewed or affected by developmental level. Additionally, the HBM does not take into account a parent’s health beliefs or other parent factors nor adolescents’ level of responsibility for their own health behaviors.
While there has been very little research specifically applying the HBM to adolescent patients, the HBM has been modified for children in the Children’s Health Belief Model (Bush & Iannotti, 1990) by adding factors from Social Learning Theory, Cognitive Development Theory, and Behavioral Intention Theory. The adapted model includes modifying factors, readiness factors, and behavior factors. The modifying factors include cognitive/affective factors (health locus of control, self-esteem, health risk-taking, medical knowledge, medical autonomy), enabling factors (physician visits, illness frequency), and environmental factors (caregiver’s motivations for the child, parental perception of illness threat, perceived benefit of medicines, and expected child medication use). The environmental factors are separated into readiness factors and behavior factors. Readiness factors include motivation, perceived illness threat, and perceived benefit of the medicines. The behavior factor was expected medication use, which was used as a predictor of actual medication use. The authors tested the adapted model on preadolescent children (aged 8-14) and their expectations of taking medicines to treat common health conditions (cold, fever, upset stomach, etc.). The authors found that their model was supported, explaining a significant portion of the variance in children’s plan to take medicine. The strongest factors included severity of illness and perceived benefit, which were both positively correlated with predicted medication use. Caregiver factors made a significant but not strong contribution to variance in children’s plan to take medicine, with the strongest factors being perceived child vulnerability and the amount of concern for their child. Although this model is one of very few models adapted for children, it has not been tested with adolescents. As identified in previous research (Bond et al., 1992), age impacts the HBM’s ability to predict adherence. Additionally, adolescents differ from children on a range of factors, including developmental level, increased independence, and motivations for behavior. Therefore, the Children’s Health Belief Model is
not sufficient for examining this population, but may be helpful in choosing important factors to study adolescent adherence.

**Previous Research on Factors Related to Adherence in SCD**

When studying adolescents, it is important to consider both adolescent and parent contributions to adherence, as measuring only parents or adolescents does not adequately account for the contributions that each makes to adherence. Research supports the importance of including adolescent psychosocial functioning, parental stress, and parental perceived barriers when examining contributions to adherence in adolescence.

Psychosocial functioning can be a product of many interacting factors in the adolescent’s life and impacts adherence to a treatment regimen. Better psychosocial functioning is related to better adherence for children and adolescents with chronic illnesses (Brownbridge & Fielding, 1994). Better psychosocial functioning also predicts adherence during a four year follow-up in adolescents with chronic illnesses (Alan et al., 1990), indicating a long-term impact of psychosocial functioning in regard to adherence. However, higher levels of psychosocial functioning are not present for all youth with SCD; rather, psychosocial comorbidities are common in this population (Anie, 2006; Panepinto, Pajewski, Foerster, Sabnis, & Hoffmann, 2009). A study of pediatric SCD patients (aged 2-18) found 40.4% of youth with SCD had some type of neurobehavioral comorbidity (anxiety, depression, attention problems, behavior problems, developmental delay, etc.), with 5.8% of the sample having anxiety problems and 7.7% having depression (Panepinto et al., 2009). Generally, research has identified similar rates of psychosocial comorbidities in healthy youth and youth with SCD (Anie, 2006; Yang, Cepeda, Price, Shah, & Mankad, 1994). However, when children and adolescents with SCD experience psychosocial problems, the consequences are more severe than for youth without SCD. For
adolescents with SCD, negative mood and stress have been associated with increased pain, increased health care use, and decreased school and social functioning (Gil et al., 2003). Adults with SCD rated lower on quality of life measures compared to both adults without chronic illness and adults with hemochromatosis (Anie, 2006). Research from many pediatric illness populations, including asthma, HIV, and diabetes (Bender, 2006; Gonzalez et al., 2008; Murphy et al., 2005; Williams et al., 2006), has found relationships between poorer psychological functioning and poorer adherence behaviors. However, no previous research has focused on examining the relationship between psychosocial functioning and adherence behavior specifically in adolescents with SCD.

There is some evidence that specific psychological diagnoses relate to adherence to treatment in different ways. Depression in an adolescent is a risk factor for nonadherence (DiMatteo, Lepper, & Croghan, 2000; Kovacs, Goldston, Obrosky, & Iyengar, 1992), and self-report of symptoms of anxiety and depression are negatively associated with adherence for adolescents with chronic illness (Mednick et al., 2010; Rosina, Crisp, & Steinbeck, 2003). However, there is some evidence that anxiety alone might increase adherence in some disease populations, such as children and adolescents with cystic fibrosis (White, Miller, Smith, & McMahon, 2009), pointing to the possibility of differential effects on adherence for individuals with different psychological diagnoses. Psychosocial factors have been suggested as an area to target in order to improve adherence among adolescents (La Greca & Mackey, 2009), therefore, it is imperative to understand their relationship to adolescent adherence.

As previously stated, adolescents share the responsibility of adherence with their parents (McQuaid et al., 2003). Parent psychosocial functioning, including stress, is related to adolescent adherence. Illness-related stress for parents of adolescents with SCD is the strongest predictor of
utilization of urgent care for the adolescent with SCD (Raphael et al., 2013). Parental stress is negatively related to child age and positively related to difficulty of a regimen for youth with diabetes (Streisand, Swift, Wickmark, Chen, & Holmes, 2005). That is, a parent’s illness-related stress level decreases as their child enters and progresses through adolescence and increases with the complexity or difficulty of a treatment regimen. Low family stress is related to better adherence in youth with SCD (Treadwell et al., 2005). Parent stress has been identified as an important target to improve adherence in SCD (Raphael et al., 2013).

In addition to parent stress, a parent’s perception of barriers to adherence has an impact on adherence among adolescents with chronic illnesses. In most families that have participated in research assessing barriers, at least one barrier to treatment adherence is reported (Ingerski, Baldassano, Denson, & Hommel, 2010). Identified barriers can include resistance to behavior change, forgetfulness, and a desire to not identify with having the disease (Rhee, Belyea, Ciurzynski, & Brasch, 2009). Lack of time to conduct medical tasks, medication side effects, and complicated disease regimen are also common barriers identified by parents and adolescents (Greenley, Stephens, Doughty, Raboin, & Kugathasan, 2010). In SCD, poverty status (Panepinto et al., 2009), filling prescriptions, remembering to take medication (Elliott et al., 2001), delayed benefits of adherence, difficulties with medication availability at pharmacies, the burden of monitoring adherence, and transportation difficulties (Thornburg et al., 2010) have been identified as reasons for non-adherence.

When considering both parent and adolescent contributions to adherence, it is vital to account for the level of responsibility both the adolescent and the parent take in the adherence regimen. Parents who consider the outcomes or consequences of their child’s illness as more severe are more likely to engage in adherence behaviors to care for their child compared to parents who
perceive less severe outcomes or consequences of the illness (Becker et al., 1978; Soliday & Hoeksel, 2000). For youth with SCD, independent youth responsibility for adherence tasks has been associated with lower adherence (Alvarez et al., 2009). Non-adherent youth with SCD share fewer responsibilities for adherence with their caregivers compared to youth with better adherence (Treadwell et al., 2005; Treadwell & Weissman, 2001). Additionally, those children and adolescents who perceive more support (from parents, friends, peers, and teachers) are more likely to increase responsibility for adherence and adherence over time (Treadwell & Weissman, 2001).

**The Present Study**

The goal of the present study was to investigate parent and adolescent factors related to adherence among adolescents with SCD. Previous research indicates that adherence leads to better health outcomes. Additionally, it is vital to consider both adolescent and parent contributions to adherence due to the changing role the parent and adolescent has in responsibility for disease management. Current research has identified factors that are empirically related to adherence in adolescents with chronic illness, including adolescent psychosocial functioning, parental stress, and parental perceived barriers. However, the research has not sufficiently examined both adolescent and parent contributions in predicting adherence. Additionally, research on psychosocial functioning has not been applied directly to adherence in adolescents with SCD.

The goal of the present study was to evaluate the contributions of adolescent psychosocial functioning, parental stress, and parental perceived barriers to adherence in adolescents with SCD. It also examined the contribution of responsibility for adherence to the relationship between adolescent psychosocial functioning and adherence.
Specific Aims and Hypotheses

Aim 1: Determine whether adolescent psychosocial functioning is related to adherence in adolescents with SCD.

**Hypothesis 1a:** Adolescent anxiety will account for a significant amount of variance in adherence scores, such that lower levels of adolescent anxiety will independently predict higher levels of adherence to sickle cell regimen.

**Hypothesis 1b:** Adolescent depression will account for a significant amount of variance in adherence scores, such that lower levels of depression will independently predict higher levels of adherence to sickle cell regimen.

Aim 2: Determine whether parental levels of stress and perceived barriers are related to adherence in adolescents with SCD.

**Hypothesis 2a:** Levels of parental stress will account for a significant amount of variance in adherence scores, such that lower levels of parental stress will independently predict higher levels of adherence.

**Hypothesis 2b:** Perceived barriers to healthcare will account for a significant amount of variance in adherence scores, such that fewer perceived barriers will independently predict higher levels of adherence.
CHAPTER 2

METHOD

Participants

Participants were 30 adolescents with SCD between the ages of 12 and 18 and their caregiver attending clinic visits at a Midwestern pediatric hospital. Participants were either African American (85.2%) or biracial (14.8%) adolescents mostly accompanied by their mother (85.2%). Most caregivers were single parents (81.5%) who ranged in age from 31 to 56 years old. Type of medical insurance and parent education were used as proxies for socioeconomic status. Most (88.0%) had Medicaid as their primary health insurance, followed by private insurance (12.0%). Most mothers (42.3%) had some college or professional school education and most fathers (59.1%) had a high school education.

Procedure

Graduate research assistants obtained consent from parents/guardians and assent from adolescents during a routine medical visit. One parent/guardian and the adolescent patient completed a packet of questionnaires at the clinic or at home and returned the packet during their next scheduled clinic appointment. For completing their questionnaire, the adolescents received a $10 gift card. A total of 40 adolescents and 39 caregivers consented to participate in the study, however one caregiver was excluded as two caregivers for the same adolescent completed packets. Of those who agreed to participate, 30 completed packets were received and included in the study.
Measures

Demographic Questionnaire

A demographic questionnaire was developed specifically for the present study (see Appendix A). This measure assessed the caregiver’s relationship to the adolescent, age, ethnicity, education background, marital status, and health insurance. This measure also assessed the adolescent’s age, grade, ethnicity, and health insurance. In the present study, SES was measured utilizing proxies of parent education and health insurance status. Adherence problems affect adolescents of all SES classes, but studies on SES and adherence in SCD are limited, with only two examining the relationship between income and youth medication adherence (Loiselle et al., 2016). One found higher income was correlated with higher medical staff rating of adolescent adherence (Barakat et al., 2002), while the other found no relationship between income and adherence (Fisak, Belkin, von Lehe, & Bansal, 2011).

Adolescent Psychosocial Functioning

Psychosocial functioning in the adolescent was assessed using the following measures:

Spence Child Anxiety Scale, Child Report and Parent Report [SCAS; (Spence, 1998)]. The SCAS was developed to assess anxiety across six domains (generalized anxiety, panic/agoraphobia, social phobia, separation anxiety, obsessive compulsive disorder, and physical injury fears). Respondents rate 38 items on a Likert scale from 0 (never) to 3 (always), (range = 0 to 114). Raw scores are translated to t scores, which have been developed for males and females between 8 and 15 years of age. A t-score of 60 or greater reflects elevated anxiety symptoms. Internal consistency is excellent (alpha = .90), and this scale has demonstrated internal and external validity with adolescents in Australia (Spence, Barrett, & Turner, 2003). The parent report version has good reliability (alpha = 0.89) and demonstrates internal and
external validity in a sample comparing children and adolescents with and without anxiety disorders (ages 6-18) (Nauta et al., 2004). While a portion of participants in this study fell outside of the original age range, the SCAS has been used in older adolescents and maintained high internal consistency (alpha = .92) and correlated with other measures of anxiety (Essau, Anastassiou-Hadjicharalambous, & Muñoz, 2011). The SCAS has been used with a chronic pain sample that included one adolescent with SCD (Eccleston, Crombez, Scotford, Clinch, & Connell, 2004).

**Center of Epidemiological Studies – Depression, Revised, Child Version [CES-D; (Radloff, 1977); CES-DR; (Eaton, Smith, Ybarra, Muntaner, & Tien, 2004)].** The CES-DR is a 20 item self-report measure of depressive symptoms experienced during the past week which participants rate on a scale from 1 (rarely or none of the time, less than 1 day) to 4 (most or all of the time, 5-7 days) (range = 20 to 80). The scale has been previously utilized with African American adolescents (Pittman & Chase-Lansdale, 2001). It has demonstrated excellent internal consistency (alpha = 0.93), and convergent and divergent validity in a primarily Caucasian (8.2% African American) sample of young undergraduate students (Van Dam & Earleywine, 2011).

**Parent Stress**

The *Pediatric Inventory for Parents [PIP;(Streisand, Braniecki, Tercyak, & Kazak, 2001)]* assessed stress experienced by parents. The PIP is a 42 item self-report inventory used to measure childhood illness-related parenting stress. Parents respond to each item on a Likert scale of frequency from 1 (never) to 5 (very often) and difficulty associated with the item from 1 (not at all) to 5 (extremely). Frequency and difficulty are summed separately on four domain scales: Communication (range = 9 to 45), Emotional functioning (range = 15 to 75), Medical Care (range = 8 to 40), and Role Function (range = 10 to 50), which can be added for an overall
frequency score (PIP-F; range = 42 to 210) and difficulty score (PIP-D; range = 42 to 210). Higher scores indicate greater frequency and levels of difficulty. It has demonstrated good to excellent internal consistency (alpha range: .80-.96), and has demonstrated construct validity as scores correlate with a measure of anxiety and parenting stress [PIP-F r = .34; PIP-D r = .29, both p < .001; (Streisand et al., 2001)]. The PIP has been used in a study of adolescents with SCD and their parents (Logan, Radcliffe, & Smith-Whitley, 2002).

Parent Perceived Barriers

The Barriers to Healthcare Scale [BHCS; (Fisak et al., 2011)] assessed barriers to healthcare perceived by parents. The BHCS is a 16 item self-report of caregiver perceptions of barriers to care in relation to SCD treatment recommendations. Each item has a 4 point Likert scale ranging from 1 (“never or rarely a problem) to 4 (“almost always a problem”), (range = 16 to 64). It has demonstrated good internal consistency (alpha = 0.87) in a study of parents of youth with SCD (Fisak et al., 2011).

Responsibility for Adherence

The Responsibilities Questionnaire (parent and patient version; Appendix B) assessed both parent and adolescent self-reported responsibility for different aspects of adherence. The Responsibilities Questionnaire is a five-item measure developed for the current study based on the Asthma Responsibilities Questionnaire (ARQ) (McQuaid et al., 2001). This version assesses who is responsible (parent vs. patient) for tasks related to sickle cell care. The parent and patient forms of this questionnaire are identical except for the wording of the questions. For example, instead of “Making sure I go to my appointments” (patient version), the parent form states “Making sure my child goes to his/her appointments.” Each item has a Likert scale, ranging from 1 (“parent(s) take responsibility all of the time”) to 5 (“child takes responsibility all of the
time”), with higher scores on the measure indicating more adolescent responsibility for adherence tasks (range = 5 to 25). The ARQ has good internal consistency for both the child (alpha = .75-.78) and parent (alpha = .84-.87) report and good convergent validity in a sample including African American adolescents (McQuaid et al., 2001). The ARQ has been used in studies of African American children and adolescents (McQuaid et al., 2003).

**Adherence**

The *Self-Care Inventory – Sickle Cell Disease* (SCI-SCD; parent and patient version; Appendix C) assessed adherence. There is currently no gold standard method for measuring adherence. Some self-report measures have been found to be well-established in pediatric conditions such as diabetes, but self-report or parent-report often leads to higher reports of adherence than more objective measures such as diary or electronic monitoring data (Quittner, Modi, Lemanek, Ievers-Landis, & Rapoff, 2008). The SCI-SCD is a 4 item adherence measure that examines four aspects of adherence (following diet, activity restriction, medication, and attendance at appointments) adapted for the current study from the Self-Care Inventory for Diabetes (LaGreca, Swales, Klemp, & Madigan, 1988). Participants respond to each aspect of adherence on a Likert scale from 1 (“never”) to 5 (“always”). Higher scores represent higher levels of adherence. For this study, an average of reported adherence across each aspect of adherence (range = 1 to 5) was used in analyses. If a certain aspect is not a part of a participant’s adherence, then they may answer “Not applicable”. If a particular aspect of adherence was not applicable to a participant, this aspect was not included in the participant’s adherence total. That is, adherence scores utilized for main analyses were an average reported for the tasks that were relevant to the participant. Parent and patient forms of the SCI-SCD are identical except for wording and can be utilized to compare reports of adherence. The measure from which the
The current instrument was adapted, the Self-Care Inventory for Diabetes (LaGreca et al., 1988), has good internal consistency [alpha = .80; (LaGreca et al., 1988; Lewin et al., 2009)], is well-established by Evidence Based Assessment Task Force criteria (Quittner et al., 2008), and correlates with a structured interview of adherence behaviors and lab measures of diabetic adherence (Lewin et al., 2009). The diabetes version of this questionnaire has been utilized in studies with African American children and adolescents (Lewin et al., 2009; Wysocki et al., 1996).

For a comprehensive list of constructs and measures, please refer to Table 1.

Table 1

<table>
<thead>
<tr>
<th>Construct</th>
<th>Measures</th>
<th>Notes</th>
</tr>
</thead>
<tbody>
<tr>
<td>Psychosocial Functioning</td>
<td>Anxiety: Spence Child Anxiety Scale, Child and Parent Report Depression: Center of Epidemiological Studies – Depression, Revised, Child Version</td>
<td>Anxiety and Depression were analyzed separately (two separate linear regressions) to determine if each condition has a different effect on adherence.</td>
</tr>
<tr>
<td>Adherence</td>
<td>Self-Care Inventory - SCD – Parent Report - Patient Report</td>
<td>Each analysis were performed using the average parent and patient report, separately.</td>
</tr>
<tr>
<td>Parent Stress</td>
<td>Pediatric Inventory for Parents</td>
<td>Frequency scores and Difficulty scores were analyzed separately (two separate linear regressions) to determine differential effects on adherence.</td>
</tr>
<tr>
<td>Parent Perceived Barriers</td>
<td>Barriers to Healthcare Scare</td>
<td></td>
</tr>
<tr>
<td>Responsibility for Adherence</td>
<td>Responsibility Questionnaire – Parent Form - Patient Form</td>
<td>Each analysis was performed using parent and child report, separately, matching with adherence reporter</td>
</tr>
<tr>
<td>Patient and Family Demographics</td>
<td>Demographic Questionnaire</td>
<td></td>
</tr>
</tbody>
</table>
Analysis Plan

Preliminary Analyses

Outliers. Data were evaluated for outliers before testing hypotheses, as linear regression assumes normality of data. Scores greater than 3.29 standard deviations were considered outliers and excluded from analysis (Tabachnick & Fidell, 2001). Two variables had a participant with a value greater than 3.29 standard deviations from the mean. For the CES-D, one participant obtained a z-score of 3.47, and for the BHCS a different participant obtained a z-score of 3.90. Analyses were conducted both with and without these participants; results were similar with and without these outliers. In a linear regression including all independent variables, the Mahalanobis distance was used with a chi-square critical value of $p < .001$ (Tabachnick & Fidell, 2001) to determine cutoffs for inclusion in analysis. The following results are reported with the outliers included, as there were no outliers identified using Mahalanobis distance and chi-square cut-offs.

Correlations. Pearson correlations (2-tailed) or $t$ tests were conducted between demographic variables (adolescent age, parental education, insurance type), and the study variables (adolescent- and parent-report of adolescent anxiety, adolescent depression, adolescent- and parent-reported adherence, parent stress – frequency, parent stress - difficulty, parent perceived barriers, and adolescent- and parent-reported responsibility for adherence). Additionally, correlations between parent and adolescent agreement on reports of adherence and responsibility for adherence were examined. Findings of correlations were utilized in addition to hypotheses to make decisions about which variables to include in final analyses.

Age. As the study includes a wide age range, age was explored as a variable with the potential of grouping some age groups together depending upon findings. Adherence is expected
to be worse for adolescents (Bucks et al., 2009; DiMatteo, 2004; Foster et al., 2001), while young adolescents and adults have better adherence (DiMatteo, 2004). Age was not conceptualized as a control variable, as distinctions across age ranges are considered important in this study. Rather, it was analyzed as an independent variable in a nonlinear regression to determine the contribution to variance that age provides.

**Primary Analyses**

**Hypothesis 1a:** Adolescent anxiety will account for a significant amount of variance in adherence scores, such that lower levels of anxiety will independently predict higher levels of adherence to sickle cell regimen.

Multiple regression was conducted to determine if adolescent anxiety independently predicts adherence in adolescents with SCD. Separate analyses were conducted for adolescent- and parent-reported anxiety and adherence. Report of anxiety was entered as the independent variable and adherence was entered as the dependent variable. Additionally, a t-score was utilized to compare mean level of adherence between a group of adolescents with elevated anxiety symptoms and a group of adolescents with non-clinical levels of anxiety.

**Hypothesis 1b:** Adolescent depression will account for a significant amount of variance in adherence scores, such that lower levels of depression will independently predict higher levels of adherence to sickle cell regimen.

Multiple regression was conducted to determine if adolescent depression independently predicts adherence in adolescents with SCD. Separate analyses were conducted for adolescent and parent reported adherence. Report of depression was entered as the independent variable and adherence was entered as the dependent variable.
**Hypothesis 2a:** Levels of parental stress will account for a significant amount of variance in adherence scores, such that lower levels of parental stress will independently predict higher levels of adherence.

A linear regression was conducted to determine if parental stress predicts adherence in adolescents with SCD. Separate analyses were conducted for Frequency and Difficulty. Parental stress was entered as the independent variable and adherence was entered as the dependent variable. These analyses of parental stress also included adolescent age as an additional predictor based upon correlations with Frequency of parental stress.

**Hypothesis 2b:** Perceived barriers to healthcare will account for a significant amount of variance in adherence scores, such that fewer perceived barriers will independently predict higher levels of adherence.

A linear regression was conducted to determine if perceived barriers predict adherence in adolescents with SCD. Perceived barriers was entered as the independent variable and adherence was entered as the dependent variable.

**Power Analysis**

G*Power (Erdfelder, Faul, & Buchner, 1996) was used to conduct power analyses for this study. Between 1 (predictor only) to 2 (predictor with 1 control variable) variables were included in the study analyses. In order to detect a medium effect size with an alpha of \( p = .05 \) and power = .80 with 1 independent variable, a sample size of 55 is required. With the same parameters and 2 independent variables, a sample size of 68 is required. However, as cited above, multiple studies have been published with smaller sample sizes. In order to detect a large effect size with an alpha of \( p = .05 \) and power = .80 with 1 independent variable, a sample size of 25 is required. For 2 independent variables with those same parameters, a sample size of 31 is
required. The obtained sample size is 30 and insufficient for many analyses, despite its similarity to previously reported samples. However, the study variables had very few outliers (2), which did not impact study findings. Despite normality of data, in order to minimize bias, bootstrapped confidence intervals were explored with 1,000 bootstrap samples in order to estimate biases and variances in order to increase robustness when working with a small sample size (Ievers-Landis, Burant, & Hazen, 2011). Results of bootstrapping indicated overall similar results. When bootstrapping revealed differences, they are noted in the results with bootstrapped confidence intervals. Confidence intervals that do not contain zero indicate significant results.
CHAPTER 3

RESULTS

Preliminary Analyses

Participants

More than half of parents (53.6%) reported their teen was non-adherent, and 32.1% of teens self-reported non-adherence on the average measure of adherence across four tasks of the SCI-SCD. The majority of parents (82%) and adolescents (72.4%) reported parents take the majority of the responsibility for adherence. About a quarter (25%) of parents reported experiencing some stress related to parenting an adolescent with a serious illness, and 22.2% of parents reported significant difficulty coping with parenting stress related to parenting an adolescent with a serious illness. Only 10.7% of parents reported never or rarely experiencing barriers to healthcare. A quarter of the adolescents met clinical cutoff criteria for depression (24.1%). For anxiety, 17.2% of adolescents self-reported clinical levels of anxiety and 21.4% of parents reported clinical levels of anxiety in their teenager. Descriptive statistics for the sample are listed in Table 2.
Table 2

Sample Characteristics (N = 30)

<table>
<thead>
<tr>
<th>Variable</th>
<th>M (SD) or %</th>
</tr>
</thead>
<tbody>
<tr>
<td>Adolescent age</td>
<td>14.4 (2.02)</td>
</tr>
<tr>
<td>Adolescent gender</td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>46.7</td>
</tr>
<tr>
<td>Female</td>
<td>53.3</td>
</tr>
<tr>
<td>Adolescent Ethnicity</td>
<td></td>
</tr>
<tr>
<td>African American</td>
<td>85.2</td>
</tr>
<tr>
<td>Biracial</td>
<td>14.8</td>
</tr>
<tr>
<td>Caregiver Relationship to Adolescent</td>
<td></td>
</tr>
<tr>
<td>Mother</td>
<td>85.2</td>
</tr>
<tr>
<td>Father</td>
<td>3.7</td>
</tr>
<tr>
<td>Grandmother</td>
<td>3.7</td>
</tr>
<tr>
<td>Legal Guardian</td>
<td>7.4</td>
</tr>
<tr>
<td>Single Parent Status</td>
<td>81.5</td>
</tr>
<tr>
<td>Caregiver age</td>
<td>39.32 (7.52)</td>
</tr>
<tr>
<td>Adolescent medical insurance</td>
<td></td>
</tr>
<tr>
<td>Public</td>
<td>88.0</td>
</tr>
<tr>
<td>Private</td>
<td>12.0</td>
</tr>
<tr>
<td>Mother education</td>
<td></td>
</tr>
<tr>
<td>Some high school</td>
<td>11.5</td>
</tr>
<tr>
<td>High school or GED</td>
<td>19.2</td>
</tr>
<tr>
<td>Some college/professional school</td>
<td>42.3</td>
</tr>
<tr>
<td>Associates/Bachelor’s Degree</td>
<td>26.9</td>
</tr>
<tr>
<td>Graduate/Advanced Degree</td>
<td>0</td>
</tr>
<tr>
<td>Father education</td>
<td></td>
</tr>
<tr>
<td>Some high school</td>
<td>4.5</td>
</tr>
<tr>
<td>High school or GED</td>
<td>59.1</td>
</tr>
<tr>
<td>Some college/professional school</td>
<td>22.7</td>
</tr>
<tr>
<td>Associates/Bachelor’s Degree</td>
<td>4.5</td>
</tr>
<tr>
<td>Graduate/Advanced Degree</td>
<td>9.1</td>
</tr>
<tr>
<td>Responsibility</td>
<td></td>
</tr>
<tr>
<td>Parent report</td>
<td>9.48 (4.22)</td>
</tr>
<tr>
<td>Adolescent report</td>
<td>10.66 (5.54)</td>
</tr>
<tr>
<td>Adolescent Anxiety</td>
<td></td>
</tr>
<tr>
<td>Parent report</td>
<td>16.96 (11.22)</td>
</tr>
<tr>
<td>Adolescent report</td>
<td>22.93 (13.83)</td>
</tr>
<tr>
<td>Depression</td>
<td>11.97 (8.38)</td>
</tr>
<tr>
<td>Adherence</td>
<td></td>
</tr>
<tr>
<td>Parent report</td>
<td>3.97 (.72)</td>
</tr>
<tr>
<td>Adolescent report</td>
<td>4.22 (6.13)</td>
</tr>
<tr>
<td>Parent stress</td>
<td></td>
</tr>
<tr>
<td>Frequency</td>
<td>102.22 (26.95)</td>
</tr>
<tr>
<td>Difficulty</td>
<td>95.46 (27.98)</td>
</tr>
<tr>
<td>Barriers to healthcare</td>
<td>21.74 (6.33)</td>
</tr>
</tbody>
</table>
Missing Data

Missing data were handled using listwise deletion in each analysis in order to maintain sufficient sample size for analyses. One parent packet did not have a corresponding adolescent packet due to the adolescent failing to turn in the initial packet. Three adolescent packets did not have corresponding parent packets. Two of these adolescent packets were completed by adolescents aged 18 who chose not to have their parents participate and the other parent failed to turn in the initial packet.

Preliminary Examination of Study Variables

Results from independent samples t-tests indicate a significant difference in responsibility for adherence tasks between publically insured participants and privately insured participants. Adolescents with private insurance (M = 16.0, SD = 5.0) reported significantly more adolescent responsibility for adherence tasks compared to adolescents with public insurance (M = 9.38, SD = 4.50), t(22) = -2.358, p = .028, d = 1.39. Parents of adolescents with private insurance (M = 15.33, SD = .58) reported their adolescents took significantly more responsibility for adherence tasks compared to parents of adolescents with public insurance (M = 8.55, SD = 3.89), t(23) = -2.966, p = .007, d = 2.44. Adolescents of parents with private insurance (M = 16.0, SD = 5.0) reported significantly more responsibility for adherence tasks compared to adolescents of parents with public insurance (M = 9.18, SD = 4.49), t(23) = -2.44, p = .023, d = 1.44. Parents with private insurance (M = 15.75, SD = .96) reported their adolescents took significantly more responsibility for adherence tasks compared to parents with public insurance (M = 8.41, SD = 3.62), t(19.78) = -8.08, p < .001, d = 2.77. The overall difficulty experienced related to parenting stress (PIP) approached significance (p = .093), with privately insured parents (M = 105.50, SD = 9.15) reporting slightly higher difficulty with parenting stress than publically insured parents.
(M = 91.57, SD = 29.05). No other significant nor trending differences were identified by adolescent insurance type (all ps>.10).

Chi square tests were unable to be performed comparing insurance type and parent levels of education, as 75%-90% of expected cell counts were below 5 for all comparisons. Three patients in the sample had private insurance; all of these families reported mother’s education level as an Associate’s or Bachelor’s Degree and father’s education as high school or graduate degree. For the publically insured patients, both mother’s and father’s education ranged from some high school through Associate’s or Bachelor’s degree. For the publically insured patients, both mother’s and father’s education ranged from some high school through Associate’s or Bachelor’s degree. Four parents in the sample reported private insurance, with mother’s level of education at some college or an Associate’s or Bachelor’s degree. Privately insured parents reported father’s level of education as a high school education or a graduate degree. For parents with public insurance, both mother’s and father’s level of education ranged from some high school through Associate’s or Bachelor’s Degree.

Correlations results revealed adolescent age was negatively correlated with parent stress – Frequency (r = -.38, p < .05). Father’s level of education (r = .46, p < .05) was positively correlated with adolescent-report of responsibility for adherence. Adolescent-reported adherence was positively correlated with parent-reported adherence (r = .53, p = .006). Notably, parent- and adolescent-report of responsibility for adherence were not correlated (p =.121). Additionally, parenting stress – Frequency was positively correlated with parent-reported barriers to healthcare (r = .54, p < .01). This correlation makes conceptual sense, as an increase in barriers to healthcare would logically lead to more frequent experiences of parenting stress. No changes were made to analyses based upon this expected correlation. Correlations are presented in Table 3. In addition to these correlations, bootstrapped confidence intervals found correlations between age and parenting stress – Difficulty (r = -.41, 95% CI [-.742, -.036]). Mothers’
education was significantly correlated with parent report of responsibility for adherence (a trend discovered before bootstrapping data; $r = .46$, 95% CI [.114, .736]).

Table 3

**Bivariate Correlations between Study Variables (N = 29)**

<table>
<thead>
<tr>
<th>Variable</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
<th>8</th>
<th>9</th>
<th>10</th>
<th>11</th>
<th>12</th>
<th>13</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Age (A)</td>
<td>-</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. Parent education (M)</td>
<td>.001</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. Parent education (F)</td>
<td>.001</td>
<td>.46*</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. Anxiety (A)</td>
<td>.14</td>
<td>.30</td>
<td>.42*</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Anxiety (P)</td>
<td>-.17</td>
<td>.12</td>
<td>.04</td>
<td>.68***</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Depression (A)</td>
<td>.33*</td>
<td>.17</td>
<td>.36</td>
<td>.60***</td>
<td>.49*</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>7. Adherence (A)</td>
<td>-.17</td>
<td>-.11</td>
<td>.19</td>
<td>.29</td>
<td>.15</td>
<td>.14</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>8. Adherence (P)</td>
<td>-.07</td>
<td>.18</td>
<td>.20</td>
<td>.11</td>
<td>-.02</td>
<td>.09</td>
<td>.53**</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>9. Parent stress - Frequency</td>
<td>-.38*</td>
<td>.21</td>
<td>-.04</td>
<td>.19</td>
<td>.47*</td>
<td>.01</td>
<td>.27</td>
<td>.17</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>10. Parent stress - Difficulty</td>
<td>-.04</td>
<td>.14</td>
<td>.03</td>
<td>-.06</td>
<td>.19</td>
<td>.04</td>
<td>.004</td>
<td>.10</td>
<td>.78***</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>11. Barriers to healthcare</td>
<td>-.22</td>
<td>-.17</td>
<td>.07</td>
<td>.17</td>
<td>.46*</td>
<td>.26</td>
<td>.13</td>
<td>.20</td>
<td>.54**</td>
<td>.34*</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>12. Responsibility for adherence (A)</td>
<td>.23</td>
<td>.28</td>
<td>.46*</td>
<td>.30</td>
<td>-.10</td>
<td>.27</td>
<td>.05</td>
<td>.22</td>
<td>-.05</td>
<td>-.18</td>
<td>.08</td>
<td></td>
<td></td>
</tr>
<tr>
<td>13. Responsibility for adherence (P)</td>
<td>.19</td>
<td>.34*</td>
<td>.35</td>
<td>-.01</td>
<td>-.15</td>
<td>.15</td>
<td>-.08</td>
<td>.003</td>
<td>-.03</td>
<td>.24</td>
<td>-.11</td>
<td>.31</td>
<td></td>
</tr>
</tbody>
</table>

*Note: *p < .05, **p < .01, ***p<.001, t = trending relationship, p falls between .05 and .10*

Controls

Based upon above correlations, father’s level of education was identified as a potential control for analyses including responsibility for adherence. However, responsibility for adherence was not related to adolescent anxiety, adolescent depression, nor adherence.

Therefore, responsibility (and the demographic variable related to it) was not included in the analyses for Aim 1 to optimize power in the small sample size. All other potential control variables were not related to other study variables, and they were excluded from the analysis in order to optimize power in a small sample size.
Age

Age was analyzed as an independent variable in two separate nonlinear (quadratic) regressions for parent- and adolescent-reported adherence to determine the contribution to variance that age provided. The regression was not significant for either parent-reported ($p = .850$) or adolescent-reported adherence ($p = .463$). The effect of age on adherence was also measured by comparing means of both parent- and adolescent-report of adherence across three age groups utilizing a one-way-ANOVA. Results revealed no significant differences, between any age groups, in parent report of adherence ($p = .930$) nor adolescent report of adherence ($p = .648$).

Agreement between Parent-Report and Adolescent-Report

Agreement between parent-report and adolescent-report was examined utilizing a paired-samples t-test and a sign test (Siegel, 1956).

**Adherence.** The paired samples t-test comparing the mean of parent-report of adherence ($M = 3.99$, $SD = .74$) and adolescent-report of adherence ($M = 4.26$, $SD = .58$) was not statistically significant ($p = .052$), indicating parent- and adolescent-report did not significantly differ. An exact sign test to examine agreement was also not significant ($p = .286$), indicating the medians did not vary significantly. Bootstrapping results revealed a significant difference between parent ($M = 3.99$, $SD = .74$) and adolescent ($M = 4.26$, $SD = .58$) reports of adherence (CI $[.013, .530]$). Therefore, the parent and adolescent reports of adherence were considered statistically different.

**Responsibility for adherence.** The paired samples t-test comparing the mean of parent-report of responsibility for adherence ($M = 9.19$, $SD = 4.02$) and adolescent-report of responsibility for adherence ($M = 9.92$, $SD = 4.90$) was not significant ($p = .487$), indicating
parent and adolescent mean report of responsibility for adherence did not significantly differ. An exact sign test utilized to examine agreement was not significant ($p = .503$), indicating the medians did not vary significantly.

Parent- and adolescent-report were then examined qualitatively. Responsibility was originally rated on a scale from 1-5; for ease of interpretation, responses of 1 or 2 were considered “parent is mostly responsible”, 3 was considered “responsibility is equally shared” and 4 or 5 was considered as “adolescent is mostly responsible.” Overall, parents and adolescents tend to agree that parents are mostly responsible for most tasks (see Table 4). When specific tasks were examined for each patient, there were four instances identified in which parents gave adolescents more responsibility than the adolescent claimed for that adherence task (see Figures 1-5). The tasks for which responsibility was not clear included adherence to activities they are/are not supposed to do, making sure the adolescent has medicine, and taking medicine. These patients were identified as “high risk patients” because report from both parent and adolescent indicate responsibility is not clearly assigned for these tasks, with the parent reporting the adolescent is mostly responsible and the adolescent reporting that the parent is mostly responsible.
Table 4

*Response Frequencies for Responsibility Items*

<table>
<thead>
<tr>
<th>Responsibility Item</th>
<th>Parent</th>
<th>Shared</th>
<th>Teen</th>
</tr>
</thead>
<tbody>
<tr>
<td>Teen report:</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Making sure I follow my diet</td>
<td>46.2%</td>
<td>15.4%</td>
<td>38.5%</td>
</tr>
<tr>
<td>Making sure I don't do activities I'm not supposed to do</td>
<td>52.4%</td>
<td>28.6%</td>
<td>19.0%</td>
</tr>
<tr>
<td>Making sure I have enough medicine</td>
<td>46.4%</td>
<td>21.4%</td>
<td>32.1%</td>
</tr>
<tr>
<td>Making sure I take my medication</td>
<td>44.8%</td>
<td>17.2%</td>
<td>37.9%</td>
</tr>
<tr>
<td>Making sure I go to my appointments</td>
<td>69.0%</td>
<td>17.2%</td>
<td>13.8%</td>
</tr>
<tr>
<td>Parent report:</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Making sure my child follows his/her diet</td>
<td>55.6%</td>
<td>33.3%</td>
<td>11.1%</td>
</tr>
<tr>
<td>Making sure my child doesn't do activities s/he is not supposed to do</td>
<td>59.1%</td>
<td>22.7%</td>
<td>18.2%</td>
</tr>
<tr>
<td>Making sure my child has enough medicine</td>
<td>66.7%</td>
<td>22.2%</td>
<td>11.1%</td>
</tr>
<tr>
<td>Making sure my child takes his/her medication</td>
<td>51.9%</td>
<td>33.3%</td>
<td>14.8%</td>
</tr>
<tr>
<td>Making sure my child goes to his/her appointments</td>
<td>85.2%</td>
<td>14.8%</td>
<td>0.0%</td>
</tr>
</tbody>
</table>

*Figure 1. Responsibility for Diet-Related Adherence Tasks*
Figure 2. Responsibility for Activity-Related Adherence Tasks

Note: Orange oval indicates patient at higher risk for nonadherence.

Figure 3. Responsibility for Having Medication

Note: Orange ovals indicate patients at higher risk for nonadherence.
Figure 4. Responsibility for Taking Medication

Note: Orange oval indicates patient at higher risk for nonadherence.

Figure 5. Responsibility for Attending Appointments
Adolescent anxiety. Parent- and adolescent-report of adolescent anxiety required utilizing z-scores as the range of the scales was not the same. The raw scores of the scales were significantly correlated (r = .68, p < .001), indicating these scales are related.

Hypothesis Testing

Hypothesis 1a: Adolescent anxiety will account for a significant amount of variance in adherence scores, such that lower levels of adolescent anxiety will independently predict higher levels of adherence to sickle cell regimen.

Four separate linear regressions with anxiety predicting adherence were conducted. There were no statistically significant relationships. The first linear regression utilized adolescent self-report of anxiety to predict adolescent-report of adherence (p = .133). The second utilized parent-report of adolescent anxiety to predict parent-reported adherence (p = .930). The third utilized adolescent self-report of anxiety to predict parent-report of adherence (p = .600). The fourth utilized parent-report of adolescent anxiety to predict adolescent-report of adherence (p = .472).

Based on the results of the linear regressions, we decided to explore the relationship between adolescent anxiety and adherence utilizing clinical cut-offs for anxiety. Clinical cut-off scores on the SCAS are 20.1 for boys and 21.7 for girls on the parent measure of anxiety and 33 for boys and 38 for girls on the adolescent measure of anxiety. These scores represent a t-score of 60, or the cutoff for clinical elevation on the measure. Mean differences of adolescent-report of adherence between adolescents above and below the clinical cutoff for anxiety were examined using an independent sample t-test. Adolescents above (M = 4.64, SD = .28) the clinical cutoff for anxiety reported significantly higher adherence than adolescents below (M = 4.13, SD = .63) the clinical cutoff for anxiety, t(14.34) = -2.82, p = .013, d = 1.05.

As multiple adolescents met criteria for comorbid anxiety and depression, a one-way
ANOVA was performed to examine differences in mean adherence among those high in anxiety, those high in depression, and those high in both anxiety and depression rather than a t-test examining only adolescents high in anxiety compared to adolescents high in depression. No significant differences were found in adolescent-reported adherence ($p = .475$) nor parent-reported adherence ($p = .985$) among the means for adolescents high in anxiety, depression, or both.

**Hypothesis 1b: Adolescent depression will account for a significant amount of variance in adherence scores, such that lower levels of depression will independently predict higher levels of adherence to sickle cell regimen.**

Two separate linear regressions with depression predicting adherence were conducted. There were no statistically significant relationships. The first linear regression included utilizing adolescent self-report of depression to predict adolescent-report of adherence ($p = .468$). The second utilized adolescent self-report of depression to predict parent-report of adherence ($p = .727$).

Based on the results of the linear regressions, we made the decision to further explore the relationship between depression and adherence utilizing clinical cut-offs for depression. A score of 16 on the adolescent measure of depression indicates a clinical cutoff for depression. An independent sample t-test comparing the mean of adolescent self-report of adherence for adolescents above and below the clinical cutoff was not significant ($p = .644$).

**Hypothesis 2a: Levels of parental stress will account for a significant amount of variance in adherence scores, such that lower levels of parental stress will independently predict higher levels of adherence.**
Four separate linear regressions with parent stress predicting adherence were conducted. For those examining frequency of parent stress, age was included as a predictor based upon correlations. There were no statistically significant relationships. The first linear regression utilized parent report of frequency of stress and adolescent age to predict parent report of adherence ($p = .693$). The second utilized parent report of difficulty of stress to predict parent report of adherence ($p = .632$). The third utilized parent report of frequency of stress and adolescent age to predict adolescent report of adherence ($p = .390$). The fourth utilized parent report of difficulty of stress to predict adolescent report of adherence ($p = .986$).

To explore further based upon correlations with age and parenting stress, we examined the relationship to parenting stress among older versus younger adolescents. When only including participants over the age of 14, overall difficulty associated with parenting stress was significantly related to average parent-reported adherence ($p = .043$). See Table 5 for results.

Table 5

*Summary of Multiple Regression Analysis for Parenting Stress and Parent-reported Adherence among Older Adolescents (N = 12)*

<table>
<thead>
<tr>
<th>Variable</th>
<th>B</th>
<th>SE (B)</th>
<th>$\beta$</th>
<th>$t$</th>
<th>Sig. ($p$)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Constant</td>
<td>5.62</td>
<td>.63</td>
<td>8.90</td>
<td>.000</td>
<td></td>
</tr>
<tr>
<td>Parenting Stress - Difficulty</td>
<td>-.02</td>
<td>.01</td>
<td>-.68</td>
<td>-2.46</td>
<td>.043</td>
</tr>
</tbody>
</table>

Note. $R^2 = .46$

Bootstrapping results revealed the same relationship between parenting stress and parent-reported adherence within the full sample. Overall difficulty associated with parenting stress was significantly related to average parent-reported adherence (CI [-.049, -.005]). See Table 6 for results.
Table 6

Summary of Multiple Regression Analysis for Parenting Stress and Parent-reported Adherence – Bootstrapped Results

<table>
<thead>
<tr>
<th>Variable</th>
<th>B</th>
<th>Bias</th>
<th>SE (B)</th>
<th>95% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Upper</td>
</tr>
<tr>
<td>Constant</td>
<td>5.57</td>
<td>.332</td>
<td>1.65</td>
<td>4.773</td>
</tr>
<tr>
<td>Parenting Stress - Difficulty</td>
<td>-.015</td>
<td>-.003</td>
<td>.017</td>
<td>-.049</td>
</tr>
</tbody>
</table>

Note. $R^2 = .44$

**Hypothesis 2b:** Perceived barriers to healthcare will account for a significant amount of variance in adherence scores, such that fewer perceived barriers will independently predict higher levels of adherence.

Two separate linear regressions with barriers to healthcare predicting adherence were conducted. There were no statistically significant relationships. The first linear regression utilized parent report of barriers to healthcare to predict parent report of adherence ($p = .330$). The second utilized parent report of barriers to healthcare to predict adolescent report of adherence ($p = .522$).

The most frequent barriers that participants identified as experiencing often or always were insurance/financial problems, transportation problems, time off work or school, not enough time, could not find babysitting for other children, and forgetting (see Table 7). These frequently-experienced barriers were more closely examined, as socioeconomic variables (insurance status and parent education) are theoretically related to pragmatic resource variables. Significant differences were identified when examining parent report of barriers by both parent’s insurance status and adolescent’s insurance status. An independent samples t-test indicated parents with public insurance ($M = 1.50, SD = 0.74$) indicated forgetting was a more frequently experienced barrier than parents with private insurance ($M = 1.00, SD = 0.00$), $t(21.0) = 3.17$, $p = .005$, $d =$
.96. Similarly, parents of patients with public insurance (M = 1.41, SD = 0.67) indicated forgetting was a more frequently experienced barrier than parents of patients with private insurance (M = 1.00, SD = 0.00), t(21.0) = 2.88, p = .009, d = 0.87. Additionally, there was a trend for parents of patients with private insurance (M = 2.67, SD = 1.53) to reported more frequent experiences with insurance or financial problems than parents of patients with public insurance (M = 1.64, SD = .85), p = .084. No other frequently-experienced barriers were related to insurance status (all ps > .10). A one-way ANOVA revealed a significant effect of father’s education level on frequency of experiencing transportation problems [F(2,19) = 4.92, p = .019, \( \eta^2 = 0.341 \)]. Specifically, 34.1% of the variance in transportation problems is attributable to the differences in father education level. Post hoc comparisons using the Tukey HSD test indicated the mean score for fathers with some college (M = 2.80, SD = .84) was higher than for fathers with a high school education (M = 1.65, SD = .93, p = .046) or fathers with some graduate training (M = 1.0, SD = .00, p = .025). No other frequently-experienced barriers were related to father or mother level of education (all ps > .10).
Table 7

Response Frequencies for Barriers to Healthcare

<table>
<thead>
<tr>
<th>Barrier</th>
<th>% Parents Endorsing</th>
</tr>
</thead>
<tbody>
<tr>
<td>Transportation problems</td>
<td>23.3</td>
</tr>
<tr>
<td>Insurance or financial problems</td>
<td>22.2</td>
</tr>
<tr>
<td>Could not take time off work or school</td>
<td>14.8</td>
</tr>
<tr>
<td>Could not find babysitting for other children in the family</td>
<td>10.0</td>
</tr>
<tr>
<td>Not enough time in the day</td>
<td>10.0</td>
</tr>
<tr>
<td>Forgetting about appointments, to take medication, to drink enough water, etc.</td>
<td>10.0</td>
</tr>
<tr>
<td>Concerned that treatment, medication, or other recommendations might be harmful</td>
<td>3.7</td>
</tr>
<tr>
<td>Difficulty getting my child's medication</td>
<td>3.7</td>
</tr>
<tr>
<td>Unhappy with the relationship with my child's doctor</td>
<td>3.7</td>
</tr>
<tr>
<td>Treatment is not consistent with my religious beliefs or practices</td>
<td>3.7</td>
</tr>
<tr>
<td>My child was too sick to go to appointments or follow medical advice</td>
<td>3.3</td>
</tr>
<tr>
<td>The doctor was unclear about what he/she wanted my child to do</td>
<td>3.3</td>
</tr>
<tr>
<td>Ran out of medication</td>
<td>3.3</td>
</tr>
<tr>
<td>Did not see the benefit of making the appointment, taking a medication, or following medical advice</td>
<td>3.3</td>
</tr>
<tr>
<td>Unhappy with the medical care provided to my child</td>
<td>0.0</td>
</tr>
<tr>
<td>Did not have a telephone</td>
<td>0.0</td>
</tr>
</tbody>
</table>

Note. Percentages indicate parents who responded with “often” or “always” regarding how often the barrier was a problem.

Exploratory Analyses

Initial correlations indicated relationships exist among responsibility for adherence tasks and father’s level of education. This relationship was further examined by comparing report of responsibility for adherence tasks by father level of education. Due to low frequencies of some categories, the education levels were combined into High School (some high school and high school/GED), Some College/Professional School, and Degree (Associate’s/Bachelor’s degree, some graduate school, and graduate/advanced degree). No significant differences were found
utilizing a one-way ANOVA comparing means of adolescent-reported responsibility (p = .137) and parent-reported responsibility (p = .490) by father’s level of education. When report of responsibility for adherence was examined by mother’s level of education, no education levels needed to be combined, as there were sufficient frequencies in all but one education level (none with graduate/advanced degree). Results of a one-way ANOVA revealed a significant effect of mother’s level of education on adolescent-reported responsibility for adherence tasks [F(3,21) = 4.68, p = .012, $\eta^2 = 0.4004$]. Specifically, 40.04% of the variance in adolescent-reported responsibility for adherence tasks is attributable to the differences in mother’s education level. Post hoc comparisons used the Games-Howell test, as the data did not meet assumption of homogeneity of variances. Results indicated adolescents whose mothers had a high school education or GED (M = 4.6, SD = .55) reported significantly lower responsibility for adherence-related tasks than adolescents whose mothers had some college or professional school education (M = 9.8, SD = 4.13, p = .014) and those whose mothers had earned an Associate’s or Bachelor’s Degree (M = 13.14, SD = 4.95, p = .015). See results in Figure 6.
Figure 6. Responsibility for Adherence by Mother’s Level of Education

Note: Higher levels of responsibility indicate greater adolescent responsibility for adherence tasks.
CHAPTER 4

DISCUSSION

This study examined the relationship between adherence and adolescent psychopathology, parenting stress, and barriers to healthcare in a sample of adolescents with SCD and their parents. Overall, findings did not support hypotheses, but important relationships were revealed for this understudied population. Despite limitations to the current study, the findings suggest clinical implications and future directions for research.

Summary of Findings

The main findings from this study relate to adherence in youth with SCD and fall into three main areas: adolescent anxiety, responsibility for adherence, and healthcare barriers. Adolescents high in anxiety self-reported higher mean levels of adherence than non-anxious adolescents, indicating anxiety may be a protective factor for adherence. Additionally, parenting stress was related to poorer adherence among adolescents. In this sample, parents took the majority of responsibility for a range of adherence tasks; however, there were some important tasks for which no one took clear responsibility. Barriers to healthcare in this sample were low overall, but indicated two areas for improvement (pragmatic resources and reminders). The current study extends the literature by examining the relationship between the adolescent’s psychosocial functioning and adherence, which has not been examined in adolescents with SCD. Additionally, this study is unique in examining both parent and adolescent factors related to adherence; previous research in adherence (such as studies with the HBM) has traditionally examined only one part of the dyad (parent or adolescent) and its contribution to adherence.
Results from this study provide practical contributions to the literature by identifying specific targets for improving adherence, including discussing responsibility for adherence and removing specific barriers.

**Anxiety and Adherence**

In this study, adolescents high in anxiety self-reported higher mean levels of adherence than non-anxious adolescents. While better psychosocial functioning is typically related to better adherence (DiMatteo et al., 2000; Kovacs et al., 1992; Mednick et al., 2010; Rosina et al., 2003), it is possible that the anxiety in this group was not at levels of clinical impairment and instead resulted in a helpful level of self-reminding and rule-following that resulted in better adherence. This finding is similar to previous findings of higher anxiety and higher adherence in adolescents with cystic fibrosis (White et al., 2009). However, it is possible that the higher level of self-reported adherence is related to demand characteristics and the anxious adolescent’s desire to be perceived positively. In the informed consent and assent of this study, the confidentiality of responses and the lack of impact on the quality of their medical care were emphasized. However, it is possible that the fact data collection was conducted in the context of a clinic visit made it difficult for participants to separate their answers from their anticipation of reactions of their healthcare providers. Particularly when considering anxiety and the potential for these participants to be highly driven to be perceived positively, future research should examine the relationship between anxiety and adherence in more detail. Potential areas for research should compare clinical levels of anxiety, slightly elevated anxiety, and non-anxious adolescents’ levels of adherence. In response to the limitation of demand characteristics, it is important to conduct research within a context that the participants feel certain their responses will not be judged or affect their medical care. Researchers at Cincinnati Children’s Hospital have established a Sickle
Cell Research Day in an attempt to address this problem. Participants and families are recruited in a location outside the hospital and in the context of an education day that provides information to families from healthcare and community partners. Another potential way to address these concerns is to collect data in the context of the hospital visit but after all healthcare has taken place and the patient will no longer have contact with providers. One separate limitation of the finding of higher adherence in adolescents with higher anxiety is the lack of objective evidence that these anxious adolescents are more adherent to any part of their medication regimen, as all data relied on self-report. Objective measures of adherence would help decrease the potential impact of demand characteristics on this finding. If objective measures were utilized, report of actual adherence may relate to anxiety in the hypothesized way, with greater anxiety related to poorer adherence.

Clinically, it is difficult to respond to the finding that more anxious adolescents reported higher adherence. Clinicians should consider the need to rely on lab values or other objective measures of adherence in anxious youth rather than relying on self-report. It is possible that the patient’s desire to be perceived positively makes his or her report of adherence artificially high. If the reported adherence is assumed to be an accurate report, then clinicians can identify elevated levels of anxiety as a protective factor for adherence. In this case, clinicians should counsel families that traits of anxiety can be helpful in facilitating adherence, potentially by increasing the patient’s self-efficacy in adherence tasks. Alternatively, when clinicians find patients at the other end of the activation scale who are extremely calm and perhaps apathetic, the patients can be identified as at risk for poor adherence. Those patients with low activation are in need of preventative intervention if identified in the clinic prior to identifying an adherence problem.
While we did not measure parent anxiety, this study did assess parenting stress. Specifically, for older adolescents, difficulty associated with parenting a child with a chronic illness was related to parent report of adherence. Bootstrapping results revealed this relationship between difficulty associated with parenting a child with a chronic illness and parent report of adherence throughout the whole sample (not just older adolescents). Higher levels of parenting stress are related to lower levels of adherence in this study’s sample, which is in line with previous findings in youth with SCD (Treadwell et al., 2005). The present study’s results revealing a relationship between parent stress and adherence as well as age of the adolescent is both similar to and different from previous studies of youth with diabetes (Streisand et al., 2005). That study found a parent’s illness-related stress level decreases as their child enters and progresses through adolescence and increases with the complexity or difficulty of a treatment regimen. However, in the present study, this relationship was only found for the older adolescents. It is possible that the regimen differences between SCD and diabetes accounts for the differing findings. That is, it is possible that the tasks involved in managing diabetes may follow a different developmental course than the tasks involved in managing SCD. Therefore, older adolescents with SCD possibly struggle more with adherence, resulting in increased parent stress.

Regardless of the reasons for the difference in age findings in the present study and previous studies, parent stress has been identified as an important target to improve adherence in SCD (Raphael et al., 2013). Clinicians should encourage families to invest time in improving adherence in order to decrease parent stress, particularly as their adolescent with SCD ages. Future research should examine the relationship between adherence, parenting stress, and age of the adolescent longitudinally, so that the directionality of the relationship between adherence and
parenting stress can be ascertained.

**Responsibility for Adherence**

In this study, most parents and adolescents agreed that parents take the majority of responsibility across adherence tasks. This result is similar to previous research (Oliver-Carpenter, 2011) and identifies clinical implications for transition to adult care for adolescents with SCD and promoting independence in adherence. Clinicians should incorporate formalized transition care in the treatment plan for adolescents with SCD to help make a smooth transition of responsibility from parent to adolescent before the adolescent is a young adult without high levels of parental support.

In our sample, there were instances in which the parent stated that the adolescent had more responsibility than the adolescent claimed. In some of those cases, the adolescent claimed the parent was mostly or totally responsible for that task. This finding represents a mismatch in the assignment of responsibility for the task which ultimately leaves the adherence task as undone with no one assigned responsibility. This finding may indicate a “high risk” group that would be at the greatest risk for non-adherence, as responsibility was not clearly assigned for important adherence tasks. The tasks for which responsibility was not clear included adherence to activities they are/are not supposed to do, making sure the adolescent has medicine, and taking medicine. When responsibility is not taken for these tasks, adherence suffers. An alternative explanation for the adherence tasks for which no one is claiming responsibility is that communication about those tasks may be particularly poor in some families. For example, if the adolescent believes his/her parent is responsible for making sure s/he has taken his/her medicine, but the parent believes the adolescent should be responsible for this task, then no one is taking the burden of responsibility for making sure the adolescent has taken his/her medicine. Previous
literature has identified the majority of responsibility for adherence in SCD typically is held by parents (Oliver-Carpenter, 2011), and non-adherent children and adolescents are less likely to share responsibility for adherence with their parents (Treadwell & Weissman, 2001). So, ideally, parents and adolescents would share responsibility for adherence tasks in order to maintain optimal adherence levels.

The finding that some dyads exist in which no one is taking responsibility for tasks indicates a need for clinicians to ask about responsibility during appointments to encourage conversation among family members. At the very least, it is important both parties know who is responsible for health care tasks in order for them to be successfully adherent. Future research must identify these “high risk” patients earlier, before they enter adolescence. Interventions could be developed and implemented, and research would be able to assess the impact of improved responsibility on adherence over time. Alternatively, longitudinal research would be beneficial in determining how the responsibility for adherence tasks change over time, and what factors (e.g. developmental level of the adolescent, severity of disease) are considered when making the decision of when to transition.

Additional findings revealed a relationship between responsibility for adherence and SES factors. Mothers with a high school level of education had teenagers who reported lower levels of responsibility compared to adolescents whose mothers had some college experience or degree. That is, mothers with less education were taking more responsibility for their adolescent’s adherence than mothers with more education. Additionally, adolescents and parents with private insurance reported greater adolescent responsibility for adherence tasks than those with public insurance. Taken together, these findings indicate that for families of lower SES, responsibility for adherence tasks falls more to the parent than the adolescent. Alternatively, higher SES
adolescents take more responsibility for their adherence tasks, which can be related to better adherence. One possible explanation for this finding is that higher SES families have fewer barriers to encouraging responsibility in their adolescents. More highly educated mothers may independently identify the need to support independence in their adolescent’s adherence task. Alternatively, lower SES mothers may be less likely to be employed and are able to provide more support at home by taking responsibility for adherence tasks for their adolescent. There are multiple possibilities to explain this relationship; however the most likely explanation is that larger issues of SES status and resource availability impact responsibility for adherence in multiple ways and the relationships found here are one part of a larger puzzle. Future research should examine specific ways in which lower SES parents take more responsibility for adherence tasks and how that is related to actual levels of adherence.

**Barriers to Healthcare and Adherence**

In this study, parent report of barriers to healthcare was not related to parent or adolescent report of adherence. However, research on the HBM suggests perceived barriers are strong predictors of health behaviors, including adherence (Janz & Becker, 1984). As the HBM is not adequately adapted for adolescents, who share adherence tasks with their parents, it is possible assessing perceived barriers from all people sharing responsibility for adherence would explain adherence difficulties. Our study only assessed barriers for the parents, and there is currently no developed measure to assess barriers from the adolescent’s perspective in SCD. The lack of relationship between barriers and adherence was likely impacted by the overall very low report of barriers in this sample. The most frequent barriers that were experienced included insurance/financial problems, transportation problems, time off [from work], not enough time, babysitting, and forgetting. These fall broadly into a problem of pragmatic resources (time,
transportation, or financial) and forgetting. Previous research on barriers and adherence consistently identifies forgetting as a common barrier in chronic illness populations, including asthma (Rhee et al., 2009) and SCD (Elliott et al., 2001). In SCD, pragmatic resources have been identified as barriers to adherence, such as medication availability, transportation difficulties (Thornburg et al., 2010), and poverty status (Panepinto et al., 2009). In the current study, parents and patients with public insurance were more likely to report the barrier of forgetting, which may indicate a relationship between SES and barriers. Additionally, families in which the father has some amount of college education had more difficulty with transportation than families whose fathers had more or less education. It is possible those at either end of the SES spectrum are more able to predict their transportation (e.g., provided through public assistance or a very reliable personal vehicle). However, no other SES measures were related to more pragmatic barriers (e.g., financial problems, time off of work, getting a babysitter). Future research must examine the relationship between pragmatic barriers and family resources in a more detailed way (e.g., report of family income, more specific information on barriers).

For the physicians that care for these SCD patients, these patterns of results on barriers to healthcare indicate the importance of an interdisciplinary team approach including psychosocial care providers such as social workers and psychologists. Social workers can be invaluable in addressing the pragmatic needs of this population and, therefore, helping to reduce the barriers to healthcare reported. Future research should examine the impact of interdisciplinary collaboration on patient outcomes, such as clinic attendance or adherence. While forgetting is a difficult barrier to overcome, cues to action can decrease the frequency of forgetting. Cues to action can be identified and initiated through working with a psychosocial care provider or utilizing a smartphone app to remind patients to take medications. A potentially promising intervention is
Behavioral Family Systems Therapy (BFST), which incorporates improved family communication and problem-solving to target specific barriers to adherence (Robin & Foster, 1989). Results have revealed improved adherence and health outcomes in adolescents with diabetes (Wysocki et al., 2006).

**Limitations**

The findings of the present study should be interpreted in the context of several additional limitations. First, this study relied on adolescent and parent self-report as well as parent report of their adolescent's anxiety, adherence, and responsibility. However, when possible, validated measures previously used in this population were utilized to measure these constructs. Self-report of adherence is one way adherence is measured within clinical appointments, which may increase the ecological validity of these findings. That is, often an “assessment” of adherence during a medical visit relies only upon the answer to one or two questions asked by the provider, usually in a close-ended matter (e.g., “Have you been taking your medicine?”). Lab values are useful for measuring adherence to medications, but, in SCD, many aspects of adherence are behaviors and habits performed at home and nearly impossible to measure objectively.

The sample size of this study was insufficient to detect anything but a large effect size. As a result, more information is needed in order to determine if the relationships examined in this study exist in larger populations. In our sample, adolescent depression was not related to adherence despite previous research finding a relationship in other populations. With a larger sample size, a statistical relationship may be found between adherence and depression, as exists in many chronic illness populations. However, with the larger sample sizes detecting even very small effect sizes, the clinical implications of those findings may not be practically important. In the current study, effect sizes ranged from medium to large, and variance accounted for ranged
from 34% to 46%. This indicates that significant results are likely clinically meaningful for this population. Overall, the problem of small sample size exists across research studying SCD and suggests the need to combine data from multiple sites in order to obtain large enough sample sizes.

**Implications and Future Directions**

Adherence is complicated in SCD; therefore, any method of improving it would prove helpful in this population. Average adherence across four adherence tasks in this study ranged from one half to two thirds of the sample, which falls within the range of self-reported adherence from previous studies (12.5% to 96%) but below the average (66.5%) from a recent meta-analysis (Loiselle et al., 2016). Additionally, adolescent report of adherence was higher than their parent report, indicating a need to assess multiple family members for accurate understanding of adherence. Clinicians working with SCD have had trouble intervening in adherence, and few studies have been conducted examining empirically-supported treatments for adherence in this population (Walsh et al., 2014). Overall, clinicians should utilize multicomponent interventions including ongoing monitoring of adherence, assistance in preventing medication mistakes, and increased education of medication risks and benefits when working with this group.

Future research should expand the scope of this study by examining other explanations of the relationships among adherence, adolescent psychosocial comorbidities, parent stress, and barriers to healthcare. The hypotheses of this study were largely unsupported, so future research should examine other factors that contribute to adherence among adolescents with SCD. These findings could lead to the construction of a more thorough model for understanding adherence and responsibility for adherence among adolescent patients with SCD.
Future research would benefit by adding multiple methods of measuring adherence. For medication adherence, many electronic monitoring pill boxes have been successfully used in other studies. For other aspects of adherence, healthcare provider ratings of adherence could provide a perspective over the course of treating the adolescent rather than a simple cross-sectional view, similar to the assessment utilized in the current study. Additionally, any self-report measures of adherence should use multiple items developed via factor analysis in order to limit reliance on single-item measures when examining different aspects of adherence. The current study utilized four aspects of adherence to obtain an overall adherence score; however, specific adherence tasks (e.g., taking medicine) relied on a single item.

It would be beneficial to assess causality in the relationships between adherence and comorbid psychosocial diagnoses (anxiety, depression) and parent experiences (parenting stress, barriers to healthcare). A longitudinal study with a larger sample size may help to reveal the directionality of relationships among these variables, and, in turn, inform development of interventions to improve health outcomes.

Future research should address small sample size, adherence intervention development, and health outcomes related to adherence. The small sample size in the current study and many other SCD studies could be improved by involving multi-site studies. There is currently a call from the National Institutes of Health for funding the development of a Sickle Cell Disease Clinical Research Network, so, the feasibility of multi-site studies and larger sample sizes will likely improve in the future. Additionally, SCD is a population in need of interventions to improve adherence. To date, there are limited existing studies of adherence interventions, with no randomized clinical trials studying medication adherence among adolescents with SCD (Thornburg et al., 2010). It was beyond the scope of the current study to examine health
outcomes related to adherence in SCD. However, examining the effect of adherence on the patient’s experience of health (e.g., pain crises, hospitalizations, ED visits) would improve understanding of the patient’s motivation for adherence.

**Summary and Conclusion**

In summary, the present study found that adolescents high in anxiety self-reported higher levels of adherence than non-anxious adolescents. Most parents and adolescents agreed that parents take the majority of responsibility across adherence tasks for their adolescents with SCD and those with higher SES reported higher adolescent responsibility for tasks. The most frequently reported barriers to healthcare included pragmatic resources (time, transportation, or financial) and forgetting. Reports of adherence differed between parents and their adolescent, with more than half of parents reporting that their adolescent was non-adherent and only one third of teens self-reporting non-adherence. Clinical implications include many areas identified for impacting adherence within clinical settings, including identifying risk factors (lower SES families), promoting protective factors (some levels of anxiety), and working in multidisciplinary teams. Interventions to improve adherence will be complicated and multifaceted, considering the interaction of the need for support for responsibility in adherence tasks in a developmentally appropriate way and family resources and ability to support the adolescent in adherence tasks. One promising treatment option, BFST, is effective in addressing barriers to adherence for other adolescent chronic illness populations and could be implemented with adolescents with SCD. Future research should examine other aspects contributing to adherence, the health outcomes related to adherence, and interventions to improve adherence in adolescents with SCD utilizing multisite research to increase sample sizes.
REFERENCES


of type 1 diabetes adherence behaviors: The Self-Care Inventory (SCI). *Journal of pediatric psychology*, 34(9), 999-1007.


Appendix A
Demographic Form

Are you the primary caregiver for the patient being seen at Akron Children’s Hospital?
☐ Yes    ☐ No

What is your relationship to the patient?
☐ Mother    ☐ Father    ☐ Stepmother    ☐ Stepfather
☐ Grandmother    ☐ Grandfather    ☐ Legal guardian    ☐ Other: ______________________

Teen Date of Birth: ____________________ Parent Date of Birth ____________

Who currently resides in the teen’s residence?

<table>
<thead>
<tr>
<th>Relationship to Teen</th>
<th>Age</th>
<th>Relationship to Teen</th>
<th>Age</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td></td>
<td>6.</td>
<td></td>
</tr>
<tr>
<td>2.</td>
<td></td>
<td>7.</td>
<td></td>
</tr>
<tr>
<td>3.</td>
<td></td>
<td>8.</td>
<td></td>
</tr>
<tr>
<td>4.</td>
<td></td>
<td>9.</td>
<td></td>
</tr>
<tr>
<td>5.</td>
<td></td>
<td>10.</td>
<td></td>
</tr>
</tbody>
</table>

How would you describe your racial or ethnic background? (please check all that apply)
☐ African American    ☐ Asian    ☐ Caucasian    ☐ Hispanic    ☐ Native American
☐ Pacific Islander    ☐ Bi-Racial    ☐ Other    ☐ Prefer not to Respond

How would you describe your teen’s racial or ethnic background? (please check all that apply)
☐ African American    ☐ Asian    ☐ Caucasian    ☐ Hispanic    ☐ Native American
☐ Pacific Islander    ☐ Bi-Racial    ☐ Other    ☐ Prefer not to Respond

What is the mother’s (or female guardian’s) highest level of education? (if applicable)
☐ Some High School    ☐ High School or GED    ☐ Some College or Professional School
☐ Associates or Bachelors Degree    ☐ Some Graduate School    ☐ Graduate or Advanced Degree

What is the father’s (or male guardian’s) highest level of education? (if applicable)
☐ Some High School    ☐ High School or GED    ☐ Some College or Professional School
☐ Associates or Bachelors Degree    ☐ Some Graduate School    ☐ Graduate or Advanced Degree

Are you a single parent?
☐ Yes    ☐ No

What type of insurance provider do you have?
☐ Medicaid/Buckeye/Care Source/HMO    ☐ Private Insurance    ☐ Self Pay/No Insurance

What type of insurance provider does your teen have?
☐ Medicaid/Buckeye/Care Source/HMO    ☐ Private Insurance    ☐ Self Pay/No Insurance

Teen’s Grade Level: ______________
Teen’s estimated grade point average for the current grading period: ____________

Teen’s estimated grade point average for the previous year: ____________
### APPENDIX B

**RESPONSIBILITIES QUESTIONNAIRE**

Below are different tasks that your doctor may have asked you to do.

**Circle who does each of the tasks:** (If you weren’t told to do something, just check N/A.)

<table>
<thead>
<tr>
<th>Task</th>
<th>My parent(s) do it all of the time</th>
<th>My parent(s) do it most of the time</th>
<th>My parent(s) and I do it equally</th>
<th>I do it most of the time</th>
<th>I do it all of the time</th>
<th>N/A</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Making sure I follow my diet</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>☐</td>
</tr>
<tr>
<td>2. Making sure I don’t do activities that I’m not supposed to do</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>☐</td>
</tr>
<tr>
<td>3. Making sure I have enough medicine</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>☐</td>
</tr>
<tr>
<td>4. Making sure I take my medication</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>☐</td>
</tr>
<tr>
<td>5. Making sure I go to my appointments</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>☐</td>
</tr>
</tbody>
</table>
## APPENDIX C
### RESPONSIBILITIES QUESTIONNAIRE – PARENT FORM

Below are different tasks that your doctor may have asked your child to do.

Circle who does each of the tasks:  (If your child wasn’t told to do something, just check N/A.)

<table>
<thead>
<tr>
<th>Task</th>
<th>I do it all of the time</th>
<th>I do it most of the time</th>
<th>My child and I do it equally</th>
<th>My child does it most of the time</th>
<th>My child does it all of the time</th>
<th>N/A</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Making sure my child follows his/her diet</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>2. Making sure my child doesn’t do activities he/she is not</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>supposed to do</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>3. Making sure my child has enough medicine</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>4. Making sure my child takes his/her medication</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>5. Making sure my child goes to his/her appointments</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td></td>
</tr>
</tbody>
</table>
APPENDIX D
SELF-CARE INVENTORY

Please rate each of the items according to HOW OFTEN YOU FOLLOWED YOUR TREATMENT INSTRUCTIONS in the past week. Use the following scale:

1 = Never
2 = Mostly not, but once in a while
3 = About half of the time
4 = Most of the time
5 = Always
NA = Not told to do it

In the past week, how often have you followed the instructions for:

1. Following your diet 1 2 3 4 5 NA

2. Taking medicine (write in each of your meds):
   
   Medication 1: ______________ 1 2 3 4 5 NA
   
   Medication 2: ______________ 1 2 3 4 5 NA
   
   Medication 3: ______________ 1 2 3 4 5 NA
   
   Medication 4: ______________ 1 2 3 4 5 NA

3. Going to appointments 1 2 3 4 5 NA

4. Not doing activities you’re not supposed to 1 2 3 4 5 NA
## APPENDIX E

**SELF-CARE INVENTORY – PARENT FORM**

**HOW OFTEN HAS YOUR CHILD FOLLOWED HIS/HER TREATMENT INSTRUCTIONS**

*in the past week? Use the following scale:

1 = Never  
2 = Mostly not, but once in a while  
3 = About half of the time  
4 = Most of the time  
5 = Always  
NA = Not told to do it

**In the past week, how often has your child followed the instructions for:**

1. Following his/her diet

<table>
<thead>
<tr>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>NA</th>
</tr>
</thead>
</table>

2. Taking medicine  *(write in each of his/her meds):*

<table>
<thead>
<tr>
<th>Medication 1: __________</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>NA</th>
</tr>
</thead>
<tbody>
<tr>
<td>Medication 2: __________</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>NA</td>
</tr>
<tr>
<td>Medication 3: __________</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>NA</td>
</tr>
<tr>
<td>Medication 4: __________</td>
<td>1</td>
<td>2</td>
<td>3</td>
<td>4</td>
<td>5</td>
<td>NA</td>
</tr>
</tbody>
</table>

3. Going to appointments

<table>
<thead>
<tr>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>NA</th>
</tr>
</thead>
</table>

4. Not doing activities he/she is not supposed to

<table>
<thead>
<tr>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>NA</th>
</tr>
</thead>
</table>